Development of a screening tool for children with Fetal Alcohol Spectrum Disorder: A preliminary study

by

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Summary

Background: Children with Fetal Alcohol Spectrum Disorder (FASD) experience a broad range of cognitive and behavioural impairments. As a result, they are at risk of adverse long-term outcomes, including poor school outcomes, increased risk of developing mental health concerns, and contact with law enforcement. Individuals with FASD, particularly those with a lack of sentinel facial features, often go unrecognised and experience delays in recognition and in receiving support. Early screening for cognitive and behavioural impairments may enable children who are at risk of FASD to be identified and referred for follow up testing and support in a timelier manner. While several screening tools for FASD have been proposed, these existing tools either consider the cognitive and behavioural impairments associated with FASD narrowly, focusing on externalising behaviours, or rely on potentially expensive assessments and equipment that limit the ability to roll out at scale implementation.

Aims: This thesis sought to develop a screening tool, following a rigorous measure development process for children between the ages of four and 12 years old who display the cognitive and behavioural impairments consistent with having FASD. The aim was to develop a tool that is quick to administer, easy to use, and covers the breadth of cognitive and behavioural impairments experienced by children with FASD as identified in the literature and by experts including caregivers and clinicians.

Methods and results: This research used a mixed methods approach and involved three major study phases. In the first phase, two studies were conducted to identify the salient domains and behaviours for inclusion in a screening tool. Firstly, a scoping review of the literature was conducted. Secondly, interviews were conducted with caregivers raising children with a FASD diagnosis (n = 15) and clinicians with experience in the diagnosis or treatment of children with FASD (n = 5). These studies highlighted a broad and varied set of cognitive and behavioural

difficulties associated with FASD across the domains of self- regulation, adaptive functioning, and cognitive ability.

In the second phase, an initial pool of 300 items was developed, reflecting the findings of the scoping review and stakeholder interviews. This item pool was refined and a 95-item pool was reviewed by an expert panel, and then further refined by the research team, resulting in a 70-item initial version of the proposed screener for pilot testing. Preliminary reliability and validity testing was conducted among 83 caregivers of children between the ages four to 12 either with FASD or from a community sample to examine the psychometric properties of the Preliminary FASD Screening Tool (PFST). A total of 57 items were retained to form the PFST. The screening tool demonstrated adequate psychometric properties (Cronbach's α= .992, average inter-item correlation= .693, test-retest reliability= .988) and accurately differentiated between children with FASD and those without (Sensitivity= 0.976, Specificity= 0.927). The PFST was compared to an existing screening tool; the Neurobehavioural Screening Tool, and evidence for the validity of the tool was gathered by comparing the tool to existing measures of behaviour and executive functioning. The PFST demonstrated a high level of convergent validity. There was, however, a potential bias noted among the community sample as participants in this group predominantly were biological parents and reported having post-graduate education, whereas the FASD group were predominantly in care other than that of their biological parents. The overlap between FASD and experiences of childhood trauma were also highlighted.

Conclusion: Through a rigorous measure development process the PFST was developed, assessing a broad range of cognitive and behavioural characteristics salient to children with FASD. The PFST demonstrated a high level of accuracy in identifying children with FASD from the community sample. While the performance and psychometrics of the PFST appear promising, there is a need to conduct further research investigating the PFST in larger, and more diverse populations, and using a wider range of assessments for convergent/divergent validation.

Declaration

This thesis presents work carried out by myself and does not incorporate without acknowledgment any material previously submitted for a degree or diploma in any university; to the best of my knowledge it does not contain any materials previously published or written by another person except where due reference is made in the text; and all substantive contributions by others to the work presented, including jointly authored publications, are clearly acknowledged.



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Chapter 1 Introduction

Children with Fetal Alcohol Spectrum Disorder (FASD) experience a broad range of cognitive and behavioural impairments. As a result, they are at risk of adverse long-term outcomes, including poor school outcomes, increased risk of developing mental health concerns, and contact with law enforcement. Individuals with FASD often go undiagnosed and experience delays in the recognition of the brain-based nature of their difficulties and consequently also experience delays in receiving appropriate support. Early screening for cognitive and behavioural impairments may enable children who are at risk of FASD to be identified and referred for follow up testing and support in a timelier manner.

To support the earlier identification of children who may have FASD, the aim of this thesis is to develop a screening tool for children between the ages of four and 12 years old who may be at risk of having FASD. The intention is to develop a screening tool that is quick and easy to administer, can be easily understood and completed by caregivers of children with FASD, and covers the breadth of cognitive and behavioural impairments experienced by children with FASD, as identified in the literature and by experts. Such a screening tool could support early identification, assessment, referral, and support for children who may have FASD or other cognitive and behavioural impairments. The present chapter provides an overview of FASD and some of the challenges to identifying children living with FASD. This chapter will then review existing FASD screening tools and assess their strengths and limitations as screening tools for this population, before providing an outline of the measure development methodology adopted by the thesis.

FASD Diagnostic Approaches

The constellation of symptoms seen in children heavily prenatally exposed to alcohol was originally described in the medical literature by Lemoine and colleagues (1968). In 1973 the term

Fetal Alcohol Syndrome (FAS) was coined by Jones and colleagues (1973) to describe the symptoms seen in children born to mothers with alcoholism: namely growth impairments, facial dysmorphology, and damage to the Central Nervous System (CNS). FASD has since been adopted to capture the range of effects that can occur following Prenatal Alcohol Exposure (PAE). When first described, FASD was a non-diagnostic term encompassing a range of disorders all of which had alcohol use as an etiological factor[1]. More recently, there has been a shift in the terminology to recognise FASD as a diagnostic term with two subtypes: FASD with three sentinel facial features (i.e., small palpebral fissures, a smooth philtrum, and a thin upper lip), and FASD with less than three sentinel facial features, as reflected in the Australian, Canadian, and Scottish diagnostic guidelines[2] (Bower & Elliott, 2016; J. L. Cook et al., 2016; Scottish Intercollegiate Guidelines Network, 2019).

A diagnosis of FASD is made considering three criteria: 1) maternal alcohol use and other exposures during pregnancy, 2) assessment of the sentinel facial features, and 3) assessment of neurodevelopmental impairment. The neurodevelopmental assessment is the lengthiest aspect of the diagnostic process. Ten domains of cognitive and behavioural functioning are stipulated and ideally all ten would be assessed when a diagnosis of FASD is being formulated. The ten domains are: 1) Brain structure/neurology, 2) Motor skills, 3) Cognition, 4) Language, 5) Academic achievement, 6) Memory, 7) Attention, 8) Executive functioning including impulse control and hyperactivity, 9) Affect regulation, and 10) Adaptive behaviour, social skills or social communication. A diagnosis of FASD requires that severe impairment, defined as either in the 3rd percentile or more than two standard deviations below the mean, be demonstrated in at least three

[1] The disorders under the FASD umbrella included: FAS, Partial Fetal Alcohol Syndrome (pFAS), Alcohol Related Neurodevelopmental Disorder (ARND), Alcohol Related Birth Defects (ARBD) and Static Encephalopathy: Alcohol Exposed (SE:AE).

[2] Several diagnostic guidelines for FASD have been proposed including the Canadian diagnostic system (J. L. Cook et al., 2016), the Australian diagnostic guide (Bower & Elliott, 2016), the 4-Digit System (Astley, 2000), and Hoyme's clarification of the Institute of Medicine criteria (Hoyme et al., 2005).

of 10 domains (Bower & Elliott, 2016). The Australian FASD Diagnostic guidelines (Bower & Elliott, 2016) state that FASD should, ideally, be diagnosed by a multidisciplinary team including, but not limited to, a paediatrician, psychologist, speech and language pathologist, and an occupational therapist. Alternatively, assessments of the various domains may be conducted with a range of service providers, with one clinician coordinating all assessments.

Growth restriction (e.g., low weight and height for age) has previously been included in the diagnosis of FASD but more recent guidelines have removed growth restriction as a diagnostic criterion. The growth restriction criterion was removed as it is considered neither sensitive nor sufficiently specific for FASD diagnosis. The Australian guidelines do state, however, that growth should still be assessed as a routine element of any paediatric examination (Bower & Elliott, 2016).

Diagnostic tests intended to provide a definitive answer on whether someone has a particular condition such as FASD, can be complex, expensive, and require sophisticated analysis and interpretation (Evans, Galen, & Britt, 2005). This complexity is evident in the diagnosis of FASD: where a diagnostic assessment ideally requires input from multiple clinicians with assessments occurring over extended time frames and costs extending into the thousands. One Canadian study estimated that the time required to complete one diagnostic evaluation was between 32 to 47 hours, and cost between CA$3000 and CA$4500 (Popova, Lange, Burd, & Rehm, 2015). Similarly, a UK study describes cases assessed at the National FASD Clinic as receiving 12 hours of direct testing over two days and a similar amount of time for report writing (Mukherjee, Cook, Norgate, & Price, 2018).

Prevalence of FASD

The prevalence of FASD has been estimated in active case ascertainment studies at between two and five percent of the general population of school-aged children in the United States (May et al., 2018), Canada (Popova, Lange, Poznyak, et al., 2019), and Italy (May et al.,

2006). A study of screening prevalence based on the presence of cognitive and behavioural impairment and PAE in a large birth cohort has estimated that as many as 17% of the UK general population are at risk of having FASD (McQuire et al., 2019). Currently there are no accurate prevalence estimates for the Australian general population. Available prevalence estimates have been based on passive surveillance methods and have found rates between 0.01 and 0.68 per 1,000 births. These figures are thought to significantly underestimate the true prevalence of FASD among the Australian population (Burns, Breen, Bower, O' Leary, & Elliott, 2013). Higher prevalence rates have been found in specific populations including: the Lililwan study in the Fitzroy Valley which found the prevalence of FASD to be 19.4% (194.4 per 1,000, 95% CI = 131.0-279.0) (Fitzpatrick et al., 2017), and the Banksia Hill Youth Detention study where the prevalence of FASD was 36% (Bower et al., 2018). However, these populations are not representative of the general population.

Impact of FASD on Children and Families

Exposure to alcohol prenatally disrupts brain development (for a review of mechanisms see Sulik (2014)), and has been shown to result in damage to the brain. Overall brain size is reduced as are regions including frontal lobes, parietal lobes, and the corpus callosum. In addition to changes in size, there is also evidence of different patterns of brain activation when conducting higher order cognitive tasks (Nuñez, Roussotte, & Sowell, 2011). As a result of the diffuse damage caused by PAE, individuals with FASD can present with impairments in general intelligence, motor skills, attention, executive functioning, language, learning and memory, visual-spatial abilities, adaptive functioning, and academic performance (Mattson, Bernes, & Doyle, 2019). The cognitive and behavioural difficulties experienced by children with FASD put them at an increased risk of adverse longer-term outcomes, including poor school outcomes, contact with youth justice, poor mental health, and alcohol or other drug problems (Streissguth et al., 2004). Individuals with FASD also experience a range of co-morbid physical health problems such as hypertension and

impaired cardiovascular, renal, and auto-immune functioning (J. C. Cook, Lynch, & Coles, 2019; Popova et al., 2016; Reid, Akison, Hoy, & Moritz, 2019; Reid, Moritz, & Akison, 2019). Moreover, FASD is a lifelong condition, and adults with FASD also report increased behaviour problems, greater difficulty in paying attention, learning, memory, and in analysing social situations (Moore & Riley, 2015). Further adults with FASD report experiencing health problems such as auto-immune disorders (e.g., rheumatoid arthritis), chronic ear infections and hearing loss, and thyroid disorder persisting throughout their life (Himmelreich, Lutke, & Travis, 2017).

Benefits of Early Identification

Early identification, coupled with treatment, has been shown to improve outcomes, enhance functioning, and prevent compounding impairments/secondary conditions for a number of neurodevelopmental disorders including developmental delay and for autism (Marlow et al., 2019). Early FASD diagnosis has been shown to reduce the risks of some poorer long-term outcomes. Diagnosis after age 12, however, has been associated with an increased risk of disrupted schooling (e.g., suspension, expulsion, or dropping out), trouble with the law (e.g., being charged, arrested, convicted or otherwise in trouble with the law), confinement (e.g., time spent in juvenile detention, jail, prison, or a psychiatric hospital) and alcohol/drug problems (e.g., receiving treatment for drug problems) (Streissguth et al., 2004). Qualitative studies with parents and carers of children with FASD suggested delayed diagnosis is a significant barrier to the prevention of secondary conditions (Petrenko, Tahir, Mahoney, & Chin, 2014). An accurate FASD diagnosis aids parents and carers in understanding that their child's behaviour arises from underlying brain damage associated with PAE (Domeij et al., 2018). Furthermore, a diagnosis enables parents and caregivers to receive more accurate information and specialist advice to support their child's development (Chamberlain, Reid, Warner, Shelton, & Dawe, 2017).

Understanding the etiology of a condition contributes valuable information to understanding an individual's clinical profile. For parents and carers raising children with FASD,

understanding the brain-based nature of their children's disabilities is important for adapting the approach taken in parenting and supporting these children (Olson, Oti, Gelo, & Beck, 2009). Interventions proposed for FASD may not meet the needs of children if prenatal alcohol exposure is not recognised and typical interpretations of behaviour and parenting approaches may be a poor fit for children with FASD (Petrenko, 2015). Interventions for FASD can be improved if the underlying brain-based disability is acknowledged and if they are adapted to meet the needs of individuals with FASD (Kambeitz, Klug, Greenmeyer, Popova, & Burd, 2019; Olson, 2015; Petrenko, 2015). Effective therapies and interventions for children and families living with FASD include interventions with parents focusing on psychoeducation about FASD, and providing parents with information about behaviour management strategies, and advocacy tools (Petrenko, 2015). Interventions which seek to address self-regulation skills have also been shown to be effective among children with FASD (Reid et al., 2015).

There is also a need for additional research on pharmacological therapies for children with FASD with a recent review suggesting a lack of evidence to inform decisions about the effectiveness of medication versus potential side effects (Mela et al., 2018). Further, children with FASD may respond differently to stimulant medications that are prescribed for ADHD. Children with FASD showed a greater response to dexamphetamine but not methylphenidate (Peadon & Elliott, 2010).

Challenges and Opportunities Associated with the Early Identification of FASD

Challenges

While accurate identification is an important goal, the identification of children living with FASD presents a considerable challenge, and children are often mis-diagnosed or remain undiagnosed. A US study re-examining a random sample of 547 children previously assessed at a specialist child clinic found that 80% of the 156 children who received a diagnosis with FASD on re-examination had either been mis-diagnosed (most commonly as ADHD), or diagnosis was

missed on first presentation (Chasnoff, Wells, & King, 2015). There are a number of challenges in identifying children with FASD including lack of physical features, lack of awareness amongst clinicians about FASD, and comorbidity and overlap in presentation with other conditions.

The lack of sentinel facial features in most cases, with up to 80% of children with FASD not displaying any of the three sentinel facial features, contributes to difficulties in identifying children with FASD (Mattson, Riley, Gramling, Delis, & Jones, 1998). Several authors have suggested that children with FASD with the sentinel facial features may be identified earlier because of the physical phenotype, and this early identification may assist in accessing support services (Fagerlund, Autti-Rämö, Hoyme, Mattson, & Korkman, 2011; Streissguth et al., 2004).

A lack of FASD awareness and expertise amongst clinicians and researchers is also believed to contribute to the challenges in identifying children with FASD and hence the under-estimation of prevalence. In addition, a lack of awareness among professionals has been discussed by caregivers of children with FASD and identified as a barrier to receiving diagnostic assessments (Domeij et al., 2018; Petrenko et al., 2014). Clinician participants at training events about FASD assessment and diagnosis processes also identify access to allied health services to conduct assessments, and the length of assessment processes as barriers to undertaking FASD assessments (Reid, White, et al., 2019).

A further challenge associated with the identification of FASD is that individuals with FASD often present with several comorbid conditions. These include: Attention Deficit/Hyperactivity Disorder (ADHD), intellectual disability, and conduct disorder (Chasnoff et al., 2015; Popova et al., 2016; Weyrauch, Schwartz, Hart, Klug, & Burd, 2017). Further, children with FASD are over-represented amongst children in contact with child welfare systems and out-of-home care. (Fuchs, Burnside, Marchenski, & Mudry, 2010; Popova, Lange, Shield, Burd, & Rehm, 2019). Experiences of abuse and neglect are also associated with changes to cognitive development (Nemeroff, 2016; Teicher & Samson, 2016). Children who have experienced abuse

and neglect have been shown to have lower cognitive ability, associative learning, language development and executive functioning (e.g., working memory, inhibition, planning, and sustained attention) (McLaughlin, Sheridan, & Nelson, 2017). The overlap of the behavioural presentation of FASD and these comorbid conditions can result in children receiving diagnoses without the underlying brain damage associated with PAE being taken into account. Several different conditions and exposures may result in similar kinds of impairments: the cognitive and behavioural presentations may look similar, but the etiology may be distinct. Without specific efforts to enhance the detection and diagnosis of FASD, the etiological role of prenatal alcohol exposure may be overlooked, and the condition may go unrecognised, or misdiagnosed.

Opportunities

Children undergo rapid cognitive and behavioural development during the preschool to primary school age period, and significant developments are shown in a range of cognitive and behavioural domains including self-regulation, attention, social skills, language and memory (Diamond, 2002; Goswami, 2011). During childhood, children's development is heavily influenced by an individual's environment and their interactions with others (Sameroff, 2010). Interactions with others through processes of socialization, scaffolding, and stimulation shape emerging abilities vital for functioning including emotional regulation, and executive functioning (Fay-Stammbach, Hawes, & Meredith, 2014; Morris, Houltberg, Criss, & Bosler, 2017). Children's experiences continue to shape the development of children's brains throughout their lives. The potential for the brain to change in response to experience or environment stimulation is greatest during childhood (Farmer-Dougan & Alferink, 2013) and while adverse environments can have a significant impact during this time, interventions may take advantage of this same potential to help to address developmental concerns.

Developmental changes through childhood may also provide opportunities to identify children with FASD. Some higher order functions (e.g., executive functioning) may not emerge in

typically developing children until preschool age (Williams & Holmes, 2004). This, coupled with the increased demands on children when they start school, means that this is a time during which impairments may become more evident (Campbell, Halperin, & Sonuga-Barke, 2014). A number of cognitive and behaviour skills display age-related changes among all children and the differences between children with FASD and those without widen as children develop (Tamana, Pei, Massey, Massey, & Rasmussen, 2014; Taylor & Enns, 2017).The assessment of children at key transition times (e.g., commencing school) may assist in facilitating access to interventions (Pei, Job, Poth, & Atkinson, 2013). Identifying an individual's needs is an important driver of the subsequent recommendations for treatment (Pei, Baugh, Andrew, & Rasmussen, 2017; Petrenko et al., 2014). Given the difficulties in identifying children with FASD and barriers to diagnosis (e.g., high cost, long waiting periods), screening may assist in identifying children who are at-risk and facilitate access to further assessments, including developmental assessments and tailored supports for parents and children rather than generic neurobehavioural advice (Chamberlain et al., 2015).

A further benefit of early identification is also to support prevention efforts. If screening can facilitate population estimates of PAE and its impacts in the form of FASD, this information can assist in targeting prevention activities. An understanding of the etiology is important to guide prevention activities.

FASD Screening

Screening tools are common for many neurodevelopmental conditions including Autism Spectrum Disorders and developmental delay (Marlow, Servili, & Tomlinson, 2019), and may offer a means of identifying children who display the cognitive and behavioural functioning consistent with having FASD. Accurate screening programs can enable enhanced surveillance and access to early intervention for children who are identified as being at risk (Glover & Albers, 2007). In addition, screening instruments focusing on cognitive and behavioural functioning can provide a means for caregivers to flag concerns about behaviours, and to "sharpen" clinical

thinking about potential conditions (Frogley, John, Denton, & Querstret, 2019). Glascoe (2005, p. 174) offers the following definition of screening tools for developmental and behavioural problems: “a brief method for sorting those who probably have the problems, from those who probably do not”. Evans et al. (2005) identify that screening tools should be easy to use, cheap, and not require high levels of expertise and training to administer. Screening may enable early identification and access to support as it allows a relatively quick and inexpensive means of identifying children who display the cognitive and behavioural difficulties consistent FASD. Further, as screening does not necessarily require a skilled clinician, it can be done inexpensively and at scale. Accurate screening would ensure that the lengthy and expensive diagnostic testing is best targeted to those in the greatest need. Screening tools that can be used by non-specialists to identify those most in need of assessment may enable far larger samples including populations of children to be assessed and supported, ensuring scarce resources can be targeted appropriately (Y. Goh et al., 2008). Additionally, screening for FASD has been shown to have significant cost savings (Berrigan, Andrew, Reynolds, & Zwicker, 2019). Previous research has advocated for screening of high-risk groups for FASD to improve the numbers of children who are recognised and diagnosed, with a caveat that recognition also needs to enable access to early intervention (Petrenko et al., 2014).

In Australia, the National FASD Strategic Action Plan 2018-2028 (Department of Health, 2018) outlines the activities to be undertaken to “reduce the prevalence of FASD and the impact it has on individuals, families, carers, and communities” (p.3). The National FASD Strategic Action Plan highlights the potential of screening in both the general population and targeted screening of at-risk groups as a means of reducing the impact of FASD on individuals, families, carers and communities.

As discussed, FASD diagnosis currently relies on assessment of three core domains (i.e., facial features, cognitive and behavioural impairment, prenatal alcohol exposure). However, it is

already known that most children may not display the sentinel facial features and it is imperative that children are identified and supported regardless of the presence or absence of the sentinel facial features. In the absence of the physical features, the most salient feature of FASD is the disruption to the CNS and the associated impairments in cognition and behaviour. Despite significant research effort, at present it is unclear whether there is a consistent behavioural phenotype associated with FASD that can inform efforts towards earlier identification. A behavioural phenotype is defined as: "a characteristic pattern of motor, cognitive, linguistic, and social observations that is consistently associated with a biological disorder" (O'Brien and Yule, as cited in Kodituwakku (2007)). Identifying the common cognitive and behavioural challenges that caregivers notice in their preschool and primary school aged children with FASD could inform efforts to screen for FASD. While these challenges may not be specific to FASD, identifying common challenges could lead families and clinicians to consider potential prenatal alcohol exposure among differential diagnoses. Identifying these cognitive and behavioural challenges in preschool and primary school aged children with FASD could serve as the basis for developing screening tools to identify children who may be at risk of having FASD.

Current Cognitive and Behavioural FASD Screening Tools

Several screening tools to identify children who may have FASD have been investigated previously. Existing screening tools and methods for identifying individuals with FASD with a focus on the cognitive and behavioural impairments were reviewed[3]. Tools and methods that were not considered screening tools included:

- Studies that sought to define a FASD neurobehavioural profile, by analysing profiles of scores across neuropsychological test batteries (Kalberg et al., 2013; Lange, Shield, Rehm, Anagnostou, & Popova, 2019; Mattson et al., 2010; Mattson et al., 2013).

[3] Although identified as screening prevalence, the study by McQuire and colleagues (2019) was excluded as it presented the post-hoc screening prevalence, rather than development of a specific tool.

- Studies that sought to identify a more parsimonious set of tests when making a FASD diagnosis by examining the odds of receiving a diagnosis associated with clinically significant scores on a wide range of assessments (Enns & Taylor, 2018; Taylor & Enns, 2018).
- Screening tools that are lengthy and costly. For example, Zhang et al. (2019) who used a combination of eye-tracking and neuropsychological tests, and P. Goh et al. (2016), who proposed a decision tree involving parent and caregiver report of behavioural problems and adaptive behaviour and assessment of the sentinel facial features conducted by a paediatrician.
- Studies that have sought to screen for children with FASD using methods to assess the facial features such as digital photography (Astley, Stachowiak, Clarren, & Clausen, 2002) and 3D imaging (Suttie et al., 2013).
- Studies that have sought to use biomarkers to identify children prenatally exposed to alcohol (McQuire et al., 2016).

Three screening tools that have been applied to identify children who may have FASD on the basis of their cognitive and behavioural profile were identified: the Neurobehavioural Screening Tool (NST; Nash et al. (2006)), the Personal Behaviours Checklist-36 (PBCL-36, also known as the Fetal Alcohol Behaviour Scale; Streissguth, Bookstein, Barr, Press, and Sampson (1998)), and the Behaviour Rating Inventory of Executive Function (BRIEF; Gioia, Isquith, Guy, and Kenworthy (2000)). The NST and the PBCL-36 were developed specifically for use among individual suspected of having FASD, whereas the BRIEF was developed for a wider audience and applied to a sample of children with heavy prenatal alcohol exposure. The screening tools for the purposes of identifying FASD cases, and their potential strengths and limitations as screening tools, are summarised in Table 1.1.

Table 1.1

Summary of existing neurocognitive FASD screening tools

Screening tool: author and year	Description	Development Sample characteristics	Accuracy	Strengths	Limitations
Neurobehavioural Screening Tool (NST) (Nash et al., 2006)	A ten-item caregiver-report screening tool Developed by taking items from the Child Behavior Checklist (CBCL: (Achenbach, 1991)) which differed most between children with FASD and non-exposed peers	Nash et al., (2006) FASD (*n* = 30), ADHD (*n* = 30), CON (*n* = 30) Age range = 6-16 years, Mean age and gender split not reported	Sensitivity 63% - 98% Specificity 42% - 100% (Breiner, Nulman, & Koren, 2013; LaFrance et al., 2014; Nash, Koren, & Rovet, 2011; Nash et al., 2006)	Very brief screening tool Easy to use, and widely available Acceptable/higher levels of specificity in some samples	Focus on externalising behaviour only Lack of involvement from those with lived experience in the development of item pool
Personal Behaviours Checklist-36 /Fetal Alcohol Behavior Scale (FABS) (Streissguth et al., 1998)	A 36-item caregiver-report tool Developed from clinical experience, with a large sample of patients with FAS or FAE	Streissguth et al. (1998) *N* = 472, 46.2% female Age range = 2 - 51 years Mean not reported FAS (*n* = 169) FAE (*n* = 303)	"About 80% of the FABS scores of the normative sample fall below a score of 11 to 12, whereas about 80% of the FAS/FAE patients in the FAS/FAE reference sample have a score above 11 or 12" (Streissguth et al., 1998, p. 329)	Developed with caregiver and clinician expertise	Broad age range in development sample Despite use in subsequent research, no studies have examined either the psychometric properties or screening accuracy
Behaviour Rating Inventory of Executive Functioning (BRIEF) (Nguyen et al., 2014)	Examining the classification accuracy of the BRIEF (86 item caregiver report measure of executive function (Gioia et al., 2000)) Nguyen et al (2014) sought to examine the accuracy in identifying: children with PAE and comorbid ADHD (AE+), children with PAE without comorbid ADHD (AE-), children with ADHD and no alcohol exposure (ADHD), and a control group	Nguyen et al. (2014) *N* = 373 Age range = 8 - 16 years Mean (SD) = 12.3 (2.6) AE+ (*n* = 69) AE- (*n* = 32) ADHD (*n* = 77) CON (*n* = 141)	Overall accuracy = 71.4% AE+ = 67.1% AE- = 42.9% ADHD = 50.6% CON = 92.1% Expressed as % of groups correctly assigned through discriminant function analysis	Norms exist for comparisons to typically developing peers. Widely used measure in typically developing samples and among a range of clinical groups Strong psychometric properties	Discriminant function analysis results not easily operationalised into cut points for differentiating between groups and accuracy of the BRIEF was moderate Children who have subclinical ADHD symptoms (endorsed between 2 and 5 ADHD symptoms) from any of the study groups Use of the BRIEF is restricted and requires a licensing fee

Note. ADHD = Attention Deficit/Hyperactivity Disorder; AE+ = Alcohol Exposed, and diagnosed with ADHD; AE- = Alcohol Exposed and fewer than two ADHD symptoms; BRIEF = Behavioural Rating Inventory of Executive Function; CBCL = Child Behavioural Checklist; CON = Control group; FAE = Fetal Alcohol Effects; FAS = Fetal Alcohol Syndrome; FASD = Fetal Alcohol Spectrum Disorder; NST = Neurobehavioural Screening Tool; PBCL-36 = Personal Behaviour Checklist-36.

Neurobehavioural Screening Tool.

The NST is a 10-item proxy-report screening tool intended to identify children who may have FASD (Nash et al., 2006). Responders are asked whether their child has displayed a behaviour over the past six months, and the items of the NST are scored using a yes/no response scale. The NST is scored using an author-supplied algorithm and has scoring options to identify children who may have FASD with or without ADHD symptomatology. The NST was developed by examining items on the Child Behaviour Checklist (CBCL; (Achenbach, 1991)) that differed the most between children with FASD and their typically developing peers. In the development of the NST, 35 caregivers (e.g., biological parents and foster or adoptive parents) of children with Alcohol Related Neurodevelopmental Disorder (ARND) and 35 controls matched on age, gender, and socio-economic status completed the CBCL. A total of 62 of the 113 items of the CBCL were more frequently endorsed by caregivers of children with FASD compared to caregivers of children in the control group (Greenbaum, 2000). Twelve items were identified for inclusion in the NST based on the criterion that they "differed beyond the $p < 0.001$ level" (Nash et al., 2006, p.183). Two further items -"argues a lot" and "showing off or clowning"- were removed during testing as they were not shown to discriminate between children with FASD and/or children with ADHD or controls in a discriminant function analysis (Nash et al., 2006).

Several subsequent studies have examined whether the tool performs adequately in differentiating children with FASD from typically developing peers and other clinical populations, with mixed results. The sensitivity and specificity of the NST in identifying children with FASD has ranged between 63% to 98% and 42% to 100%, respectively (Breiner et al., 2013; LaFrance et al., 2014; Nash et al., 2011; Nash et al., 2006). The variability of the NST may be related to the variability in children's ages (i.e., 4 - 6 years old in the Breiner et al. (2013)

study, compared with 6-18 years old in the Nash et al. (2011) study), and varying cut-points and scoring algorithms used across studies. For example, Nash et al., (2011) reported 98% sensitivity and 42% specificity with a cut-point of greater than or equal to 3 items out of 10, whereas LaFrance et al., (2014) reported 63% sensitivity and 100% specificity as scored using the published algorithm.

There are several limitations of the NST to note. The NST was created from the CBCL and as such, was created from an already truncated item pool which focused primarily on externalising behaviour (Lange, Rovet, Rehm, & Popova, 2017). Being created solely from the CBCL, the NST may be lacking in construct validity as the potential item pool does not reflect the wide spectrum of difficulties experienced by individuals with FASD.

Personal Behaviour Checklist-36.

The PBCL-36- also known as the Fetal Alcohol Behaviour Scale (Streissguth et al., 1998), is a 36-item proxy score behaviour checklist developed to measure the behavioural phenotype associated with FASD. The PBCL-36 is scored using a yes/no/don't know response format. The PBCL-36 was derived from a 68-item list based on items used by parents and caretakers to describe their children with FAS, and from the first author's experience as a clinician working with families living with FAS (Streissguth et al., 1998). A reference sample of 472 individuals aged between two and 51 years old who had been diagnosed with FAS or Fetal Alcohol Effects (FAE) by dysmorphologists were used to refine the 68-item checklist to the 36-item PBCL. Items were retained in the 36-item checklist if they had an "item to scale" correlation above 0.32. The process of refining the checklist was conducted in two stages: one in 1994 and a second in 1995, and the final items represent the combination of both processes. Four age groups (2-6 years, 7-11 years, 12-17 years, and 18-51 years) were said to have been

examined separately, but no information is offered with respect to age differences across the items selected.

The ages of participants in the development sample ranged between two and 51 years old and any attempts to examine the classification accuracy have been conducted only among adult samples (Streissguth et al., 1998). Despite use in a number of studies where the PBCL differed significantly between children with FASD and typically developing peers (Kalberg et al., 2013; Kodituwakku et al., 2006; May et al., 2015; Quattlebaum & O'Connor, 2013; Viljoen et al., 2005), the psychometric properties of the PBCL have not been investigated nor its sensitivity and specificity as a screening tool. A strength of the PBCL-36 is the reflection of patient experience in the development of the initial 68 item list which served as the basis for the PBCL-36, and the clear process for reducing the number of items based on measure development guidelines.

Behaviour Rating Inventory of Executive Function.

The BRIEF was developed as a proxy-reported measure of executive functioning (Gioia et al., 2000) and consists of 86 items which generate two index scores: behavioural regulation and metacognition. The BRIEF examines a number of domains which are relevant to the cognitive and behavioural profile of children with FASD including behavioural regulation (inhibition, shifting, emotional control) and metacognition (initiate, working memory, plan/organise, organisation of materials, and monitoring task performance) (Gioia et al., 2000) Executive functioning difficulties are considered central to the behavioural functioning of children with FASD (Kodituwakku & Kodituwakku, 2014). Nguyen and colleagues (2014) examined the ability of the BRIEF to correctly classify children (n = 373) between the ages of eight and 16 years old into one of four clinical groups: PAE, defined as exposure to 14+ drinks per week, or at least weekly binge drinking, with comorbid ADHD group (n = 79), PAE without

ADHD (*n* = 36), ADHD without prenatal alcohol exposure (*n* = 90), and a typically developing control group (*n* = 168). The BRIEF accurately classified 92.1% of control subjects, 67.1% of the PAE with ADHD group, 42.9% of the PAE without ADHD, and 50.6% of the ADHD group. Nguyen and colleagues (2014) report that approximately 30% of the PAE group met the dysmorphological criteria for FAS. The BRIEF has previously demonstrated good psychometric properties among both typically developing and clinical populations such as children with ADHD and learning disabilities (Gioia, Isquith, & Roth, 2018), however the BRIEF was not developed with the purpose of classifying children with PAE from their peers and as such there may be domains which are not examined by the BRIEF which could contribute to the accuracy of screening efforts. For example, social skills are only examined in the context of behavioural regulation and there are no items relating to sleep, motor skills, or language. Further, the reported analysis presents a barrier for operationalising the findings into a screening program as cut points are not defined.

Purpose of the Thesis

The purpose of this thesis is to support screening for early identification of children who may have FASD through the development of an easy to use screening tool which builds on the body of work in this field. Previous screening tools have been developed either from pre-existing tools which were developed for other purposes, or which pre-date the current diagnostic conceptualisation of FASD and the proliferation of studies since the mid-90s which examine the cognitive and behavioural profiles of children with FASD as compared with typically developing children. This study seeks to incorporate contemporary definitions of FASD and the plethora of recent research into the development of a screening tool to identify children who display the cognitive and behavioural impairments consistent with FASD. The aim is to develop a tool that

has excellent psychometric properties, including high rates of sensitivity and specificity, and which is situated in caregiver and clinical expertise and based on the latest research findings regarding cognitive and behavioural impairments that may arise from prenatal alcohol exposure.

To support screening for early detection of children who may have FASD, the screening tool development outlined in this thesis focuses on children of preschool and primary school age. In Australia, preschool and primary school ages can span three to 13 years, however, the greatest proportion of children attend preschool and primary school between the ages of four and 12. Further, the impairments to cognitive and behavioural functioning associated with FASD may become more apparent as children make the transition to school, and the demands placed upon them increase. Transition points such as school have been highlighted as important opportunities for monitoring children's changing needs and abilities, and facilitating intervention if required (Pei et al., 2013).

The screening tool that is the focus of this thesis, has been developed according to contemporary measure development including clinician and caregiver perspectives and covers the breadth of cognitive and behavioural functioning of children with FASD. The inclusion of the patient, caregiver, and clinician perspectives is central to ensuring that the content validity (e.g., whether the instrument samples all the relevant or important content or domains) of newly developed measures is high (Brod, Tesler, & Christensen, 2009; Streiner, Norman, & Cairney, 2015). The screening tool development outlined in this thesis includes identifying the relevant cognitive and behavioural domains that a tool should aim to capture through qualitative research and a review of the extant literature, developing or adapting items that reflect these domains and having these items reviewed by experts (e.g., caregivers raising children with FASD and clinicians working in diagnostic and assessment roles), and conducting the preliminary reliability

and validity testing, including examining the accuracy of the new tool in identifying children who may have FASD (Boateng, Neilands, Frongillo, Melgar-Quiñonez, & Young, 2018; Clark & Watson, 1995; Streiner et al., 2015). Figure 1.1 outlines the methodology and chapter structure of the thesis and provides a brief summary of the content of each chapter.

Thesis Outline

Chapter 1 describes the benefits of early screening for potential FASD. The strengths and limitations of existing tools that examine the cognitive and behavioural profile with the aim of screening for FASD among children are reviewed. Chapter 2 details a scoping literature review examining the cognitive and behavioural impairment associated with PAE and FASD as compared with peers, between the ages of three and 13 years old. This literature review focuses on a broader age range than ultimately targeted in subsequent studies to ensure the breadth of cognitive and behavioural functioning is captured. In Australia, preschool and primary school ages can span 3 to 13 years, however, the greatest proportion of children attend preschool and primary school between the ages of 4 and 12 and hence, as a screening tool that may be used in these settings, this was the age range selected for testing the reliability and validity of the tool. The findings from the scoping review inform the domains of cognitive and behavioural impairment relevant for inclusion in the FASD screening tool. Chapter 3 details a qualitative study undertaken with caregivers raising children with FASD and clinicians who have expertise in the assessment, diagnosis and support of children living with FASD. Australian caregivers of children between the ages of four and 12 and clinicians participated in semi-structured interviews to examine the cognitive and behavioural difficulties experienced by children living with FASD. Thematic analysis is used to identify the most salient behaviours.

Chapter 4 describes the process of developing an initial cognitive and behavioural item pool based on a hypothesised framework generated from the findings of Chapters 2 and 3. An initial item pool is produced to capture the domains identified in the hypothesised framework. Items are developed from the content of the interviews, measures identified in the literature and pre-existing measures of concepts of interest. A panel of caregivers of children living with FASD and clinicians experienced in diagnosis and assessment review the item pool to ensure the content validity of the proposed items. The item pool is refined by the research team based on the feedback of the expert panel. Following the review process, an initial item list of 70 items is proposed for inclusion in the preliminary reliability and validity study (Chapter 5 and 6).

Chapter 5 outlines the methodology of the preliminary reliability and validity study. A preliminary reliability and validity study is undertaken with a sample of caregivers of children living with FASD and a community sample of typically developing children. Results from the preliminary study are used to refine the content of the screening tool through a quantitative item analysis process. Based on the results of this process, the 70-item screener tool is reduced to 57-items.

Chapter 6 details the results of the preliminary reliability and validity of the 57-item Preliminary FASD Screening Tool (PFST) developed through the preceding chapters. Internal consistency and test-retest reliability estimates are calculated to examine the reliability of the PFST. The validity of the tool is scrutinized. The scores of children living with FASD are compared with the community sample, and correlations with measures of related constructs such as executive functioning, behavioural concerns and trauma-related symptoms are examined along with the screening accuracy of the PFST.

Finally, Chapter 7 provides a summary of the main findings of this thesis, discussion of the implications, and directions for future research.

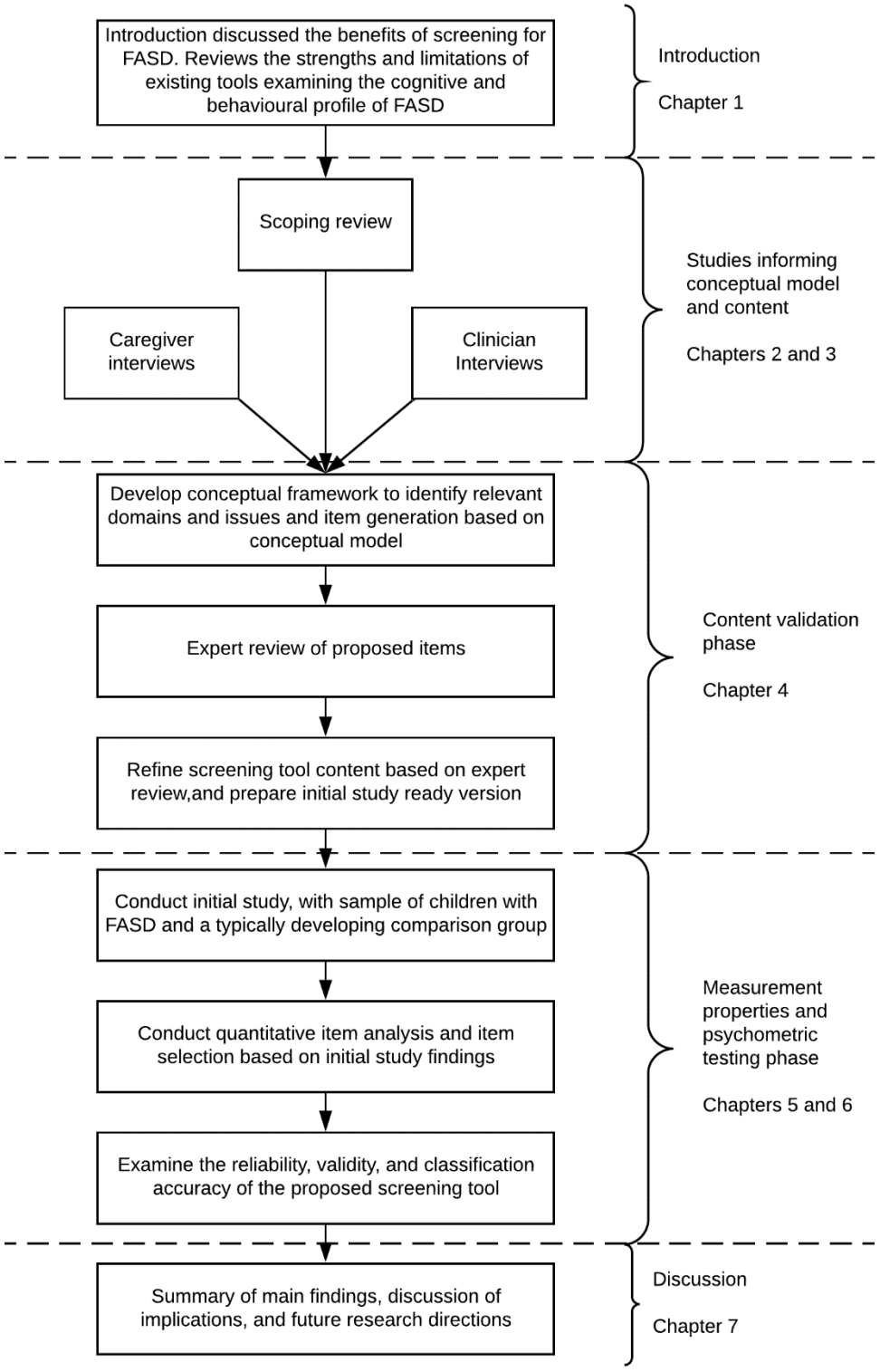


Figure 1.1. Overview of thesis methodology and thesis structure. Adapted from Brod et al. (2009).

Chapter 2 Literature Review

Preface

The previous chapter introduced the overall topics in relation to FASD and highlighted the opportunities in relation to the development of a screening tool to identify children who display the cognitive and behavioural impairments consistent with FASD. This chapter present a scoping review of existing published literature examining the cognitive and behavioural functioning of children who have either been prenatally exposed to alcohol, or who have formally been diagnosed with FASD. The results of this scoping review will be used to inform the domains and the content of a screening tool to identify who display the cognitive and behavioural impairments consistent with FASD (Boateng et al., 2018).

The literature review chapter is in the form of a scoping review. Scoping reviews follow a number of steps, similar to that of systematic reviews. However, quality assessments of included studies are not conducted Scoping reviews follow a six stage process: (1) identifying the research question, (2) identifying relevant studies, (3) study selection, (4) charting the data, (5) collating, summarising, and reporting the results, and (6) consultation phase (Arksey & O'Malley, 2005; Levac, Colquhoun, & O'Brien, 2010). Steps one through five are presented in the paper.

The scoping review has been submitted to *PLoS One*. The manuscript as submitted to the journal reflect the referencing and formatting style mandated by the journal.

Scoping Review Manuscript

Cognitive and behavioural outcomes associated with fetal alcohol spectrum disorder (FASD) and prenatal alcohol exposure (PAE): A scoping review

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Abstract

Background

Children prenatally exposed to alcohol may experience significant cognitive and behavioural impairments. Understanding the patterns of cognitive and behavioural impairments may inform efforts to identify children experiencing adverse outcomes associated with prenatal alcohol exposure early and ensure that interventions are provided.

Methods

A scoping review was conducted to examine the cognitive and behavioural impairments associated with Prenatal Alcohol Exposure (PAE), and Fetal Alcohol Spectrum Disorder (FASD) among children aged between three and 13 years old. Five databases were searched for studies that reported cognitive and behavioural outcomes among children diagnosed with FASD, or prenatally exposed to alcohol.

Results

The search yielded 6,970 results and 94 studies were included in the review. Studies were split into four methodological categories; a) studies comparing outcomes for children diagnosed with FASD, b) studies of children with PAE compared to non-exposed peers, c) longitudinal studies of the outcomes of PAE among children split into different levels of PAE, and d) longitudinal studies examining the relationship between alcohol exposure as a continuous variable and subsequent outcomes. The most frequently reported domains were executive functioning, attention, language, motor skills, and academic achievement.

Discussion

Children with PAE and FASD demonstrated impairments in a range of cognitive and behavioural domains. While consistent patterns of impairments are found across many studies, studies of children with FASD included high proportions of children who are no longer in the care of their birth parents, and the potential overlap with experiences of abuse and neglect may impact upon cognitive and behavioural functioning of children with FASD and PAE. Together with information about PAE and developmental histories of children, the patterns of cognitive and behavioural impairments identified in this study will

contribute to early identification and intervention for children with PAE/FASD to improve long term outcomes.

Introduction

Alcohol is a powerful teratogen and Prenatal Alcohol Exposure (PAE) can cause diffuse damage to the developing brain (1). PAE may result in life-long impairments across a broad range of cognitive and behavioural domains including general intelligence, executive functioning, attention, language, memory and learning, social cognition and motor functioning (2). It has been estimated that one in thirteen alcohol-exposed pregnancies results in diagnosis of fetal alcohol spectrum disorder (FASD) (3); the considerable variability in the presentation of harm following PAE is linked to differences in alcohol use patterns, gestational timing of exposure, maternal characteristics (e.g., genetics, nutrition, metabolism), and the postnatal environment (4-6). The global prevalence of alcohol use during pregnancy has been estimated at approximately 10% and varies widely across countries (7). The prevalence of FASD has been estimated at between 1% and 5% amongst school-aged children in the United States (US) (8) and Canada (9), while recent United Kingdom (UK) research estimates as many as 17% of children may have FASD (10).

Recent diagnostic guidelines developed in Australia and Canada (11, 12) have adopted FASD[4] as the overarching diagnostic term to describe cases where PAE has resulted in significant impairment in at least three cognitive and behavioural domains. These include brain structure/neurology, motor skills, cognition, language, academic achievement, memory, attention, executive function including impulse control and hyperactivity, affect regulation, and adaptive behavioural, social skills or social communication. Historically, Fetal Alcohol Syndrome (FAS) was the diagnostic term used to describe neurocognitive impairments linked to PAE, and diagnostic criteria included the presence of three sentinel facial features: a smooth philtrum; a thin vermillion border; and short palpebral fissures. Under the new guidelines, cases are

4 In the current review, FASD is used to refer to cases of FASD with less than three sentinel facial features including partial FAS (pFAS) who is a person not meeting the full diagnostic criteria for FAS but has a history of PAE, displays some facial abnormalities, as well as a growth problem or Central Nervous System abnormalities, while FAS is used to indicate the subset of children who display the three sentinel facial features.

described either as FASD with three sentinel facial features, and FASD with less than three sentinel facial features.

Many children who meet diagnostic criteria for FASD do not display the sentinel facial features associated with FAS (13, 14). The diverse cognitive behavioural presentations of children with FASD, and the lack of clinically discernible physical features makes identifying FASD challenging. In a study from the US, it was found that 86.5% of children reassessed at a specialist behavioural clinic who received a FASD diagnosis had been previously misdiagnosed, or the diagnosis was missed at their first assessment (15). Further, children who do not show the characteristic facial features experience poorer long-term outcomes than children who do, arguably as a result of the systematic barriers to accessing care in the absence of a clear diagnosis (16-18). The early and accurate identification of the cognitive and behavioural impairments associated with PAE is protective, reducing risks of poor long-term outcomes such as poor school attainment, contact with the justice system, and alcohol and other drug problems (16). To improve early identification and management of children affected by PAE, further insight into the cognitive and behavioural profiles associated with PAE and FASD in younger children is required (19).

FASD diagnostic guidelines recommend assessment of ten domains of cognitive and behavioural impairment including motor skills, cognition, language, academic achievement, memory, attention, executive function including impulse control and hyperactivity, affect regulation, and adaptive behavioural, social skills or social communication (11, 12). Previous reviews exploring the cognitive and behavioural profile of children and adolescents with FASD, FAS and/or PAE have either focused on one of these domains (e.g., 20, 21) or have not reported a systematic search strategy (e.g., 22). The aim of the current review was to systematically and comprehensively review evidence for cognitive and behavioural impairments associated with PAE, FAS and FASD in young children (19). Understanding patterns of deficits demonstrated by young children may improve the understanding of cognitive and behaviour profiles associated with PAE, FAS and FASD and contribute to early screening and interventions for affected individuals.

A scoping review methodology was used to systematically examine cognitive and behavioural domains associated with PAE in childhood, using evidence from studies of PAE, FASD, and FAS. The

review was developed to address the following question: how do young children with FASD, FAS and PAE perform in cognitive and behavioural domains, relative to typically developing TD (non-exposed) peers. This review included studies of children between the ages of three and 13, to identify patterns of cognitive and behavioural functioning that will support opportunities for earlier screening, identification and intervention.

Materials and methods

The methodology for this review was guided by the recently published Preferred Reporting Items for Systematic reviews and Meta-Analyses extension for Scoping Reviews (PRISMA-ScR) checklist and additional guidance for conducting scoping reviews (23-25).

Eligibility criteria

Table 1 shows inclusion and exclusion criteria for the review. Studies were included that reported cognitive and behavioural outcomes in children aged three to 13 years with PAE, or a diagnosis on the FASD spectrum, relative to unexposed peers. Studies with an age range of participants either above or below this range were excluded. Studies published before 1996 were excluded. This cut-off date was established as it was the publication date of the Institute of Medicine (IoM) criteria for diagnosis of FASD, and reflects the shift towards the broader definition of FASD (26). Prior to this, the term Fetal Alcohol Effects was widely used, but has since been abandoned over a lack of diagnostic clarity (27).

Table 2.1

Inclusion and exclusion criteria

Inclusion Criteria
Participant age range between 3 and 13 Published in English Published since 1996 Studies comparing the cognitive and behavioural outcomes of children with FASD, FAS, and/or PAE in cognitive and behavioural domains, relative to typically developing (non-exposed) peers, and, where possible, relative to each other
Exclusion Criteria
Animal studies No comparison group of non-exposed peers Not quantitative Use of brain imaging, or physiological assessments to assess differences
Note. FAS = Fetal Alcohol Syndrome, FASD = Fetal Alcohol Spectrum Disorder, PAE = Prenatal Alcohol Exposure

Information sources

A comprehensive, systematic search of the literature published between 1996 and 2018 was conducted in October 2018. The following databases were searched; Scopus, Medline, PsycINFO, ERIC, and EMCARE (formerly CINAHL). Search terms applied to the databases included *fetal alcohol syndrome, FAS, fetal alcohol spectrum disorders, FASD, prenatal alcohol exposure, alcohol related neurodevelopmental disorder, neurodevelopmental disorder alcohol exposed,* and *fetal alcohol effects*, combined with *child** using Boolean operators. In addition to electronic databases searched, reference lists of each included article were manually searched for articles also meeting the inclusion criteria.

Data charting process and data items

To chart the data, a data extraction form was developed and piloted on a subset of included studies by the first author (SM). Participant characteristics, study setting and recruitment procedures, FASD diagnostic criteria if relevant, methodology for determining PAE, cognitive and behavioural outcomes of interest, and a summary of key findings and limitations were extracted from all papers. The characteristics of included studies are reported in Supplementary Tables 1, 2, 3, and 4. The outcomes were grouped into

cognitive and behavioural domains defined in line with recent diagnostic guidelines (e.g., motor skills, language, academic achievement, memory, attention, executive function including impulse control and hyperactivity, affect regulation, and adaptive behavioural, social skills or social communication), excluding measurement of brain structure and neurology (11).

Data synthesis

Data analysis was a combination of both a descriptive numerical summary and qualitative synthesis describing patterns of findings across methodological categories and cognitive and behavioural domains (24).

Results

As shown in Figure 1, a total of 6,970 articles were returned from the initial search. After removal of duplicates, 4,190 studies were screened at the title and abstract level. A total of 268 articles were identified as potentially relevant and retained for full-text screening. Following full text screening, 94 articles were retained for descriptive numerical and qualitative synthesis. Reasons for exclusion at full text are shown in Figure 1.

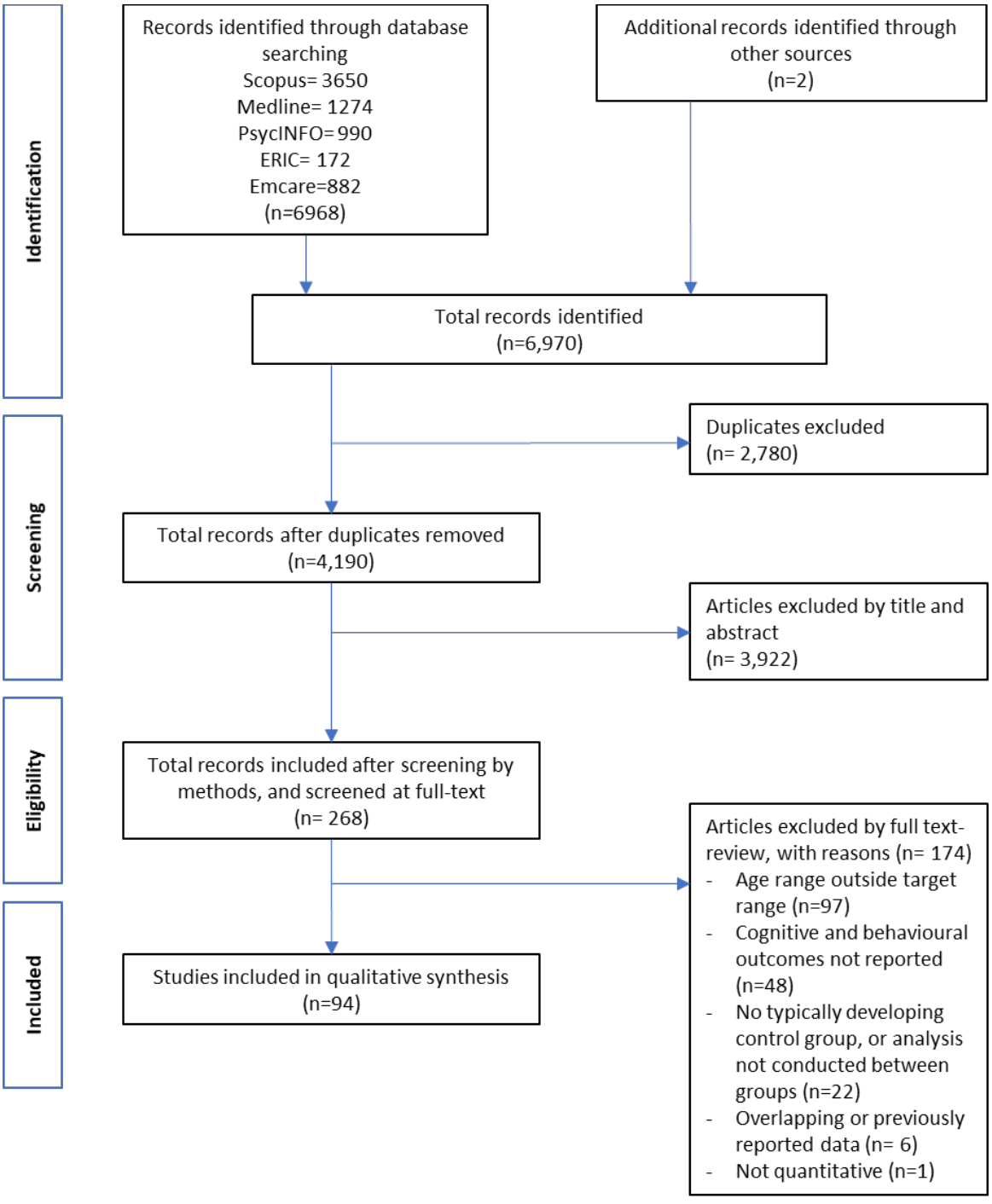


Figure 2.1 PRISMA Diagram

Studies were classified into one of four methodological categories, based on the definition of alcohol exposure, and the study design:

(1) Studies comparing cognitive and behavioural outcomes in children diagnosed with FASD to unexposed peers,
(2) Studies comparing children with PAE to unexposed peers, defined based on retrospective report of PAE, recruited where concerns about behaviour existed, or from populations believed to have a high level of alcohol exposure,
(3) Longitudinal studies of children recruited during pregnancy which compared subsequent cognitive and behavioural outcomes examined across varying categorical levels of PAE (i.e., across low, moderate, and heavy groups),
(4) Longitudinal studies of children recruited during pregnancy which examine the relationship, or lack thereof, between PAE, measured as a continuous variable (i.e., average number of drinks per day during pregnancy) and later outcomes.

Studies were split into these four groups as the methods and characterisation of alcohol exposure may have yielded different results and to allow the methodological variation within study groups to be more clearly examined. Furthermore, while studies in groups one and two typically reported differences in group mean scores using *t*-tests and ANOVA analysis, studies in group three more frequently examined the risk of outcomes such as scoring above a cut-off of clinical concern and were more frequently reported as odds ratios. Studies in group four reported either correlation or regression analyses.

Overview of included study characteristics

Of the 94 included studies, 31 studies were from the US, 19 from Canada, 14 from South Africa, eight from Australia, seven from Denmark and the UK, two from Italy, Finland and Norway, and one each from Brazil and France. The sample size of included studies ranged between 24 -114,247, and prospective cohort studies (groups three and four) tended to have much larger sample sizes. Table 2 and 3 outlines the study characteristics across the four methodological groups. For full details of the study and domain characteristics of included studies, see supplementary tables.

Table 2.2

Summary of study and domain characteristics, by methodological category

Methodological group definitions	Country of origin	Sample size range	Recruitment methods
Studies comparing cognitive and behavioural outcomes in children diagnosed with FASD to non-exposed peers;	17 Canada, 13 US, 12 South Africa, 6 Australia, 2 Italy	24 – 452	Clinic-referred (n=32), or active-case ascertainment (n=18)
Studies comparing children with PAE to non-exposed peers, defined based on retrospective report of PAE, recruited where concerns about behaviour existed, or from populations believed to have a high level of alcohol exposure,	5 US, 2 Canada, 2 South Africa	30 - 130	Clinic-referred (n=7), convenience sample in regions believed to have high PAE (n=2)
Longitudinal studies of children recruited during pregnancy which compared subsequent cognitive and behavioural outcomes examined across categorised levels of PAE (i.e., across low, moderate, and heavy groups);	7 UK, 7 Denmark, 2 Australia, 2 Finland, 1 US, 1 Brazil, 1 France	56 - 37,152	Birth cohort follow-up (n=22)
Longitudinal studies of children recruited during pregnancy which examine the relationship, or lack thereof, between PAE, measured as a continuous variable (i.e., average number of drinks per day during pregnancy) and later outcomes	11 US, 1 South Africa, 1 Norway	42 - 114,247	Birth cohort follow-up (n=13).

Table 2.3

Numbers of participants and studies across methodological groups, and domains

		Studies comparing children with FASD	Studies comparing children with PAE to non-exposed peers	Longitudinal studies comparing alcohol exposure levels	Longitudinal studies examining alcohol exposure as a continuous variable	Total
Academic Achievement	*k* Studies	7	2	6	1	16
	n Participants	687	125	24,371		25,183
	Age range	4-8.6	7-12	4-11		4-12
Adaptive Behaviour	*k* Studies	7	1	0	0	8
	n Participants	835	65	-	-	900
	Age range	5-13	6-13	-	-	5-13
Affect Regulation	*k* Studies	5	1	5	1	12
	n Participants	537	130	40670	42	41379
	Age range	6-13	6-12	5-8	5	5-12
Attention	*k* Studies	11	1	4	6	22
	n Participants	2,016	69	1756	125,888	129,729
	Age range	6-13		5-11	4-11	4-12
Executive Functioning	*k* Studies	21	4	12	4	41
	n Participants	3,426	237	58,740	9708	72111
	Age range	4-13	7-13	3-11	4-11	3-13
Language	*k* Studies	17	1	2	0	20
	n Participants	2986	51	232	-	3,269
	Age range	4-13	3-5	4-9	-	4-13
Memory	*k* Studies	5	1	2	2	10
	n Participants	399	56	128	750	1,333
	Age range	5-12	7-12	5-11	9-10	5-12
Motor Skills	*k* Studies	12	0	4	1	17
	n Participants	933	-	7816	593	9,342
	Age range	5-10	-	4-9	10	4-10
Social skills and social cognition	*k* Studies	10	3	0	0	13
	n Participants	491	142	-	-	633
	Age range	4-12	3-11	-	-	3-12
Sleep	*k* Studies	2	0	1	0	3
	n Participants	482	-	89	-	571
	Age range	3-13	-	8	-	3-13
Sensory processing	*k* Studies	3	0	0	0	3
	n Participants	105	-	-	-	105
	Age range	3-11	-	-	-	3-11

Note. PAE: Prenatal Alcohol Exposure, FASD: Fetal Alcohol Spectrum Disorders. k- Number of studies, *n*- number of participants

Studies comparing children diagnosed with FASD to unexposed peers

Of the 50 studies comparing children diagnosed with FASD to unexposed peers, there were several studies which reported different outcomes for the same cohort.[5] Six studies were reported using data collected as part of the Lililwan Study which was conducted in a remote Australian Aboriginal community (for overview of methods see Fitzpatrick, Elliott (28)).

FASD was diagnosed by the FASD 4-Digit-Diagnostic Code in 16 studies, the Revised IoM criteria in 16 studies, and a combination of the both 2005 Canadian Guidelines and the FASD 4-Digit-Diagnostic in five studies. The 2005 Canadian guidelines alone were used in three studies, while an adaptation of the 2005 Canadian guidelines was used in six studies. One study used the 1996 IoM criteria and one study used the 1989 Research Society of Alcohol guidelines (29). Two studies reported criteria other than published or adapted guidelines. Greenbaum and colleagues (30) reported using criteria established in a 2002 paper wherein a diagnosis of Alcohol Related Neurodevelopmental Disorder was assigned based on endorsement of 60% of a list of 21 deficits associated with PAE, and 50% of a list of six relative strengths (For full details of the diagnostic criteria applied in this study see (31)). Coles and colleagues (32) assigned children to a group labelled FAS/Fetal Alcohol Effects based on the scores of a dysmorphology (physical features) exam at birth and follow up. Most studies defined FASD broadly and treated diagnostic subcategories as a homogenous group. Four studies were limited to children diagnosed with FAS (29, 33-35) while five studies included only children with FAS or pFAS (36-40). Outcomes for children in FASD diagnostic subgroups (i.e., FAS, pFAS, Alcohol Related Neurodevelopment Disorder [ARND]) were compared in seven studies (38-44). FAS and pFAS groups were compared in seven studies, with ARND included in three studies. In addition to the studies comparing diagnostic subgroups, eight studies included a comparison group of children with PAE but not diagnosed with FASD (32, 36-38, 40, 41, 44-47).

Children diagnosed with FASD were frequently reported be in the care of guardians other than their birth parents. Twenty-seven studies reported the proportion of children diagnosed with FASD in the care of

5 Several studies were identified as potentially reflecting substantially overlapping samples ((1)Jirikowic, Olson et al., 2008 and Jirikowic, Kartin et al., 2008 (2); Kooistra et al., 2009, Kooistra et al., 2010, and Kooistra et al., 2011, and (3) Stevens et al., 2013, Stevens et al., 2015, Stevens et al., 2017). Efforts were made to contact the first author to confirm whether the sample overlapped.

guardians other than their birth parents and this proportion ranged between 17.5% to 100%. Twenty-three studies do not describe the caregivers of children with FASD or mention over-representation without providing data. Smaller proportions of children in care were noted in studies conducted in South Africa compared to studies from Canada and the US. Lastly, child abuse history is explicitly discussed in one study, where experience of child abuse is included as a co-variate.

Studies comparing children with PAE to non-exposed peers

Nine studies were identified comparing groups of children who had PAE but did not necessarily have a diagnosis of FASD, to non-exposed peers. These studies identified groups of children prenatally exposed to alcohol from clinic referred populations, or through targeted sampling of populations thought to being at high risk of PAE. Seven papers recruited children with PAE from larger studies or from groups attending behavioural support programs (48-54), and two studies recruited both alcohol exposed and non-exposed participants from regions where high levels of PAE had been previously identified (55, 56). Studies varied in the level of alcohol exposure used to define groups. Alcohol exposure varied from studies which included children in the PAE group based on caregivers responding “definitely yes” to a retrospective PAE screening question (52), to studies which included children in the PAE group if they had been “exposed to at least four drinks per occasion at least once per week, or 14 drinks per week” in utero (48-51, 53). For full details of how alcohol exposure was defined across these studies, see Supplementary Table 2.

Among the studies comparing children with PAE to non-exposed peers, seven studies reported the numbers of children who met some or all diagnostic criteria for FASD, or who had a pre-existing FASD diagnosis. In one study (54) included in this group, all children were reported to have a diagnosis of FASD, however, the analysis used caregiver report of PAE as a predictor of mental health diagnoses in a series of regression analyses. Of the remaining studies, the sample was examined against the diagnostic criteria for FASD, and the number of children prenatally exposed to alcohol and meeting the criteria was reported. Four studies conducted a dysmorphology assessment (assessment for sentinel facial features, and height and weight) of children recruited, and reported the proportion of children who met the dysmorphological criteria for FAS or pFAS (48-51, 53), and the proportion of children meeting the dysmorphological criteria (e.g., short palpebral fissures, smooth philtrum, and thin upper lip) of FAS with proportions meeting the

criteria ranging from 15% to 50%. One study reported the number of children with a caregiver-reported FASD diagnosis, finding that 60% of the alcohol-exposed group reported a pre-existing FASD diagnosis (52). Of the two studies of children with PAE to not state the number of children meeting FASD diagnostic criteria, one study reported dysmorphological features as an outcome measure, but did not use this to identify or report FASD cases (55) with information about the identification of potential FASD cases in the sample of children prenatally exposed to alcohol not being reported in one study (56). Similarly, to studies of children diagnosed with FASD, a high proportion of children with PAE were reported to be in care other than their birth parents, ranging from 26.7% to 95%. Three studies stated that “many” children with PAE were no longer in the care of their biological parents without providing a figure, and one study did not report this information.

Longitudinal studies comparing alcohol exposure levels

Twenty-two studies used prospective cohort study methods and compared groups based on categorised alcohol exposure levels. Six of these used data from the Avon Longitudinal Study of Parents and Children (ALSPAC) and seven used data from the Danish National Birth Cohort (DNBC). Studies varied in how alcohol exposure was defined; both in terms of the number of drinks used to define categories (i.e. low, moderate, high), the number of drinks used to define binge drinking (e.g., four or five drinks on one occasion), as well as the size of a standard drink (e.g., a standard drink is defined as 8gm in UK studies, 10gm in the Australian studies, and 12gm in Scandinavian studies). Timing of PAE also differed across studies, with some studies examining exposure throughout pregnancy, and others examining specific timeframes within pregnancy such as the first or third trimester. For full details of alcohol exposure definitions across studies, see Supplementary Table 3.

Longitudinal studies examining the relationship between cognitive and behavioural outcomes and alcohol exposure as a continuous variable

Thirteen studies examined the relationship between alcohol exposure as a continuous variable and subsequent cognitive and behavioural outcomes. Studies varied considerably in how alcohol exposure was defined, with exposure metrics including average alcohol consumption per day, total number of drinks per occasion, and scores on alcohol use disorder identification checklists. Similar to PAE category comparison

studies, studies varied with regards to considering alcohol exposure across trimesters. For full details of alcohol exposure definitions across studies, see Supplementary Table 4.

Outcomes reported

Outcomes were grouped together by subject domains to allow findings related to cognitive and behavioural domains to be examined across the four methodological categories. Key findings and study characteristics are summarised by methodological category in Tables 3-6. Studies typically included measures of multiple domains. For this reason, results are arranged in text, and in the table by domains.

(Insert Table 3 through 6 about here)

Academic achievement

Academic achievement is defined as skills including reading, mathematics, and literacy. Sixteen studies reported academic achievement outcomes and included scores on general academic achievement tests (e.g., providing an overall score, without breaking the score down into subject areas such as numeracy and literacy), or performance on tests of literacy, spelling, reading, or mathematics.

Seven studies compared academic achievement between children diagnosed with FASD and non-exposed peers (32, 33, 40, 57-60), all of which included children between four to eight years old. Children diagnosed with FASD performed more poorly than peers on measures of general academic achievement (57, 59) (32), mathematics (33, 40, 58, 60) and spelling (40, 58). Differences on mathematics and spelling persisted after controlling for IQ (58). Two studies reported reading outcomes with mixed findings; one study found children diagnosed with FASD had significantly lower reading abilities than non-exposed (40), while the other reported no differences between children diagnosed with FASD and non-exposed peers on reading skills, after adjusting for IQ (58).

Two studies compared children with PAE to peers on academic achievement outcomes (48, 55). Children with PAE performed more poorly on mathematics assessments than non-exposed peers at age 10 (48) and in grade one to three (55). Literacy outcomes were examined in one study which found children with PAE performed more poorly on Brigance literacy scales compared to non-exposed peers (55).

Six longitudinal studies compared academic achievement outcomes across categorised levels of PAE. Heavy PAE (defined as > 7 drinks per week during pregnancy, including binge drinking), was associated with increased risk of not achieving minimum standards in reading or writing, but not maths in Australian third-grade students (61). Similarly, exposure to at least one occasion of binge drinking in pregnancy (4+ drinks/day) was associated with lower mean scores on standardised national education testing in the ALSPAC cohort at age 11 (62). Mean scores decreased further with increasing frequency of binge drinking episodes in the ALSPAC cohort (63). When the ALSPAC cohort was reanalysed to compare low PAE (defined as <1 drink per week) to non-exposed children, no relationship was reported between low PAE and academic achievement (64). Similarly, in the UK Millennium cohort, exposure to 1-2 drinks per week was not associated with reading or maths skill deficits at age seven, when compared to non-exposed peers (65). Children exposed to at least 1.5 fluid ounces of alcohol per day (one fluid ounce = 29.57 ml, or approximately three standard drinks), however performed more poorly on mathematics assessments than non-exposed peers at age 4.5 years (66).

One study was identified examining the relationship between level of PAE and academic achievement. Alcohol exposure was defined as average alcohol consumption per day across pregnancy. Average alcohol consumption was found to be negatively related to Wide Ranging Achievement Test-Revised arithmetic, reading, and spelling at age six (67).

Adaptive behaviour

Adaptive behaviour refers to the constellation of conceptual, social, and practical skills that enable individuals to function in their everyday lives. Adaptive behaviour assessments include social skills, practical skills and conceptual skills such as language, reading, reasoning, knowledge and memory. While there is overlap with other domains, adaptive behaviour can be considered the application of these domains to everyday life and is considered as a stand-alone domain in FASD diagnostic guidelines (11, 12). Eight studies identified in this review reported adaptive behaviour outcomes and assessments of adaptive behaviour including the Vineland Adaptive Behaviour Scales (VABS), the Scale of Independent Behaviours-Revised (SIB-R), and clinician-rated personal/social skills on the Griffiths Mental Development Scales.

Seven studies compared children diagnosed with FASD to non-exposed peers (29, 33, 36, 40, 68-70). Five studies detailed that children diagnosed with FASD had poorer parent- and teacher-rated adaptive behaviour skills than non-exposed peers (29, 40, 68-70). Lower adaptive behaviours scores were noted among early school-aged children diagnosed with FASD (40, 70). Two studies from disadvantaged regions of South Africa reported mixed results on the clinician-scored Personal-Social subscales from the Griffiths Mental Development Scales. The earlier study found no differences between children diagnosed with FAS and non-exposed peers, after controlling for maternal education, between the ages of six and eight (33). In contrast, a larger subsequent study reported significantly lower adaptive functioning among children diagnosed with FAS or pFAS aged between five and six relative to non-exposed peers, however, maternal education was not controlled for in this study (36).

One study compared adaptive behaviour between children with PAE and non-exposed peers. Children aged six to 13 who were heavily exposed to alcohol prenatally (at least four drinks per occasion at least once per week, or 14 drinks per week throughout pregnancy) had lower VABS total scores, and lower socialisation, living skills, and communication subscale scores, a finding which was replicated when re-analysed using an IQ-matched subset of children with PAE (49).

Affect regulation

Affect regulation is defined under the current Australian FASD diagnostic guidelines as the presence or absence of symptoms of mood or anxiety disorders (11). For the purposes of this review, scores on internalising behaviour scales were included under the affect regulation domain. Twelve studies included in the review reported affect regulation outcomes.

One study included self-reported depressive symptoms as measured by the Pictorial Depression Scale (PDS; (71)). Children aged six to 12 diagnosed with FASD self-reported more symptoms of mood disorders compared to non-exposed peers on the PDS (69). Five studies compared affect regulation outcomes amongst children diagnosed with FASD and non-exposed peers (30, 32, 69, 72, 73) using the Child Behaviour Checklist (CBCL). Among the studies reporting internalising behaviours as measured by the CBCL, two studies reported children aged six to 12 diagnosed with FASD had greater internalising behaviour problems compared to non-exposed peers (69, 72), while three studies found no differences

between children diagnosed with FASD and non-exposed peers (30, 32, 73). Two studies finding no differences between children with FASD and peers were conducted in disadvantaged populations; predominantly African American families living in low Socio-Economic Status (SES) areas (32), and Australian Aboriginal families in remote areas (73). Further, children diagnosed with FASD had greater teacher-reported internalising behaviour problems than non-exposed peers in three studies (30, 69, 73).

Affect regulation was assessed in one study comparing children with PAE to non-exposed peers. PAE, identified through retrospective maternal reporting, was associated with symptoms of separation anxiety and generalised anxiety disorder but not depression. Symptoms of depression and anxiety were assessed using the Computerized Diagnostic Interview Schedule for Children- Fourth Edition among children aged six to 12 years old (54).

Among longitudinal studies comparing PAE levels, exposure to binge drinking at least weekly in the first six weeks of pregnancy was associated with increased risk of scoring above clinical cut-offs on the Strengths and Difficulties Questionnaire (SDQ) internalising behaviour and emotional problems scales at age six (74). In addition, heavy PAE was associated with increased risk of abnormal scores on the CBCL Internalising scale at age eight (75). Boys but not girls who were exposed to binge drinking had higher mean internalising behaviour scores among children aged seven in the DNBC (76). In contrast, binge drinking in the first 16 weeks or after the 30th week of pregnancy[6] was not associated with either an increased mean score or above average SDQ internalising scores among the DNBC (77).

In the one study to examine the correlation between alcohol exposure as a continuous variable, and subsequent outcomes, a significant, positive correlation was found between the maternal-reported number of drinks per drinking occasion during pregnancy and self-reported depressive symptomatology at age four to five, assessed using the Pictorial Depression Scale (78).

[6] Children exposed to any alcohol at any time outside either the first 16 weeks, or after the 30th week were excluded from the analysis conducted by Niclasen, Andersen, Strandberg-Larsen and colleagues

Attention

Various frameworks for understanding the components of attention are discussed in the literature, with this review drawing on the definitions of *sustained, selective*, and *switching attention* (79) to organise findings. Outcomes reported in the attention domain include both direct assessments of attention such as the Continuous Performance Test, and parent/teacher ratings of attention problems. Twenty-two studies reported attention outcomes.

Eleven studies compared children diagnosed with FASD to peers, using both direct assessments of attention (k = 5) and parent/teacher reports of attention (k = 8). Two studies reported both direct assessments and proxy- reports. Mixed results were found on direct assessments of attention. On the Conners' Continuous Performance Test (CPT), children seven to 10 years old diagnosed with FASD as compared to peers were found to make more errors of omission and have greater variability in response speed, suggesting impulsive rather than inattentive responding in one study (80). In contrast, among a sample of seven- to eight-year-old children diagnosed with FASD no differences were reported on any CPT or Vigilance task metrics. However a number of children diagnosed with FASD were unable to complete the task and only those able to complete the task were retained in the analysis (32). Children aged 11 years old diagnosed with FAS or pFAS in South Africa, were also reported to have greater sustained attention difficulties than their peers as measured by 'on-going task absorption' during a task assessing their ability to retain and follow instructions (37). In one study children diagnosed with FASD displayed lower performance in the executive control domain of attention and displayed greater difficulties than peers in detecting errors and inhibiting responses. Children diagnosed with FASD did not differ on tasks of sustained or selective attention (81). Finally, one study was included which compared children to non-exposed peers matched on mental age rather than chronological age (82). It stated children diagnosed with FASD (Mean age = 11.7) performed better than peers matched for mental age but were chronologically younger (Mean age = 8.4) on measures of selective attention, but not on measures of switching. It was noted in this study that 50% of the children diagnosed with FASD were unable to successfully complete the selective attention task.

Children diagnosed with FASD were consistently rated by parents and caregivers as having greater attention difficulties compared to non-exposed peers. Of the eight studies reporting observer-reports of attention, both parent and teacher ratings were included in five studies. Children diagnosed with FASD were rated by parents as having greater inattention than peers in three studies (32, 69, 82) which also persisted after controlling for IQ (69). Two Italian studies reporting data collected in separate waves of Active Case Ascertainment (ACA) among first-grade classrooms in the same region, reported contrasting parent-rated attention results. Aragón, Coriale (57) found no difference between children diagnosed with FASD and peers on parent-rated attention, whereas Kodituwakku, Coriale (59) reported children diagnosed with FASD as performing more poorly than peers. Teachers reported children diagnosed with FASD to have greater attention problems than peers in five ACA studies of children in the first grade (Approximately seven years old) (42-44, 57, 59). Similarly, teachers rated children in the first to third grades who were exposed to alcohol prenatally, but who had not undergone a FASD diagnosis assessment, as having greater attention problems than unexposed peers (55).

Six longitudinal studies compared attention outcomes between categorised groups of children exposed to various levels of PAE. Two studies directly tested attention (83, 84), finding children with PAE performed more poorly than non-exposed peers on the CPT at age 11 (83), and on an attention composite comprising sustained attention and a Go/No-Go task when tested between the ages of five and nine (84). In contrast, no statistically significant association between non-exposed, low (1-4 drinks/wk.), moderate (5-8 drinks/wk.) or heavy (9+ drinks/wk.) groups or binge drinking (dichotomised yes/no) and scores on the Test of Everyday Attention for Children at Five (TEACh-5) at age five were reported (85). However, when attention outcomes were dichotomised into an average group and low group made up of those children scoring < 1SD below the overall mean, heavy PAE (9+ drinks/wk.) was associated with an increased risk of being in the low group for both selective and sustained attention (86).

Five studies reported correlations between PAE measured as a continuous variable and child attention outcomes (87-92). Of these, four studies reported direct assessments of attention. One study reported negative associations between binge drinking in pregnancy and CPT scores at age seven (88). Similarly, a metric for alcohol exposure derived from maternal reports of alcohol consumption during

pregnancy and scores on a standard alcohol use screening tool were significantly related to CPT scores in four to five year old children (92). In contrast, no association between average alcohol consumption per day during pregnancy was reported for attention as measured by the CPT (91), or a focus-sustained attention composite measure comprising WISC-III Digit Cancellation and Coding, CPT, and Category fluency (87). Three studies reported correlations between observer-reported measures of attention and average alcohol consumption in pregnancy. Alcohol consumption in pregnancy was positively associated with parent-rated attention and impulsivity problems for children four to eleven years old in one study (89). In contrast, no association was found between average alcohol exposure per day and parent-rated attention at age seven (88). Lastly, higher levels of PAE as measured by AUDIT-C scores obtained shortly after delivery, were associated with greater attention problems as measured by the Conner's Parent Rating Scale but not a 6-item ADHD scale derived from the CBCL (90).

Executive functioning

Executive functioning refers to a collection of inter-related processes responsible for purposeful, goal-directed behavior (93). While numerous frameworks of executive functioning exist, this review draws on the review by Kingdon and colleagues (20) in arranging the literature into sub-domains of working memory (the capacity to temporarily hold and manipulate information in order to guide future response and behaviour), set-shifting (the ability to switch rapidly between difference response sets), inhibition (the ability to suppress a response, interrupt an ongoing response, and resist distraction from external stimuli), planning (the ability to identify and organize the steps needed to achieve a goal), and fluency (initiative, generativity, and productivity). While Kingdon and colleagues (20) included a vigilance (i.e., sustained attention) category in their review, this measure was considered under the attention domain in this review. Additionally, observer reports of executive functioning such as the Behaviour Rating Inventory of Executive Functioning (BRIEF), and observer reports of hyperactivity, impulsivity, and externalising behaviour are included in the executive functioning domain, following the definitions of the Australian FASD diagnostic guidelines (11). Executive functioning outcomes were reported in forty-one studies.

A total of 21 studies compared executive functioning in children diagnosed with FASD and non-exposed peers, and 15 of these reported direct assessments of executive functioning. Working memory

outcomes were reported in ten studies (40-44, 60, 69, 70, 94, 95). Seven studies examined working memory among first-grade students (approximately seven years old) as part of the ACA studies in the US and South Africa. Children diagnosed with FASD performed more poorly than peers on the backward digit span test in six studies (40-44, 94), and the Differential Ability Scales working memory composite in one study (70). A further three studies examined working memory among clinic-referred groups. Children diagnosed with FASD as compared with peers were reported to have poorer performance on the Working Memory Battery Test for Children digit recall, word-list recall, and non-word list recall but not the backward digit recall task (60) in one study (95). Lastly, one study found no differences between children with FASD and non-exposed peers on digit span after controlling for IQ (69).

Inhibition outcomes were reported in four studies (80, 95-97). One study found children diagnosed with FASD to perform more poorly than peers on the Go-No/Go task (80). Studies showed mixed results on the day/night task wherein children are required to say “sun” when presented with a picture of a moon and vice versa, with one study finding children to performed more poorly than peers (95) and a second study reporting no differences between children diagnosed with FASD and peers (97).

Set-shifting (defined as the ability to switch between response sets) was assessed in three studies (32, 69, 96), all of which found children diagnosed with FASD had greater difficulties with set-shifting relative to non-exposed peers (32, 69, 96). Differences in set-shifting were maintained after controlling for age (96), and IQ (69). Fluency outcomes were reported in one study which found poorer performance in the FASD group relative to non-exposed peers on both letter and category fluency tasks (34). Similarly, one study reported poorer outcomes among children diagnosed the FASD relative to non-exposed peers when assessing performance on the Rey-Osterrieth Complex Figure Test (98).

Teacher- and parent-reported executive function was included in one study, with children diagnosed with FASD rated lower than non-exposed peers on the behavioural regulation and meta-cognition indices of the BRIEF, after adjusting for IQ (69). Three studies examined observer reports of hyperactivity and impulsivity in children with FASD (57, 59, 69), with two studies reporting children diagnosed with FASD to have greater difficulties with hyperactivity and impulsivity compared to non-exposed peers (59, 69) and one study finding no difference (57). Mixed results were also noted on observer

ratings of externalising behaviour in children with FASD. Children diagnosed with FASD were rated by parents and caregivers as having greater CBCL externalising behaviour problems in three studies (30, 69, 72) but not different from non-exposed peers in two studies, with both conducted in disadvantaged populations (32, 73). Teacher-ratings were reported in three studies, with two studies reporting greater externalising behaviour difficulties amongst children diagnosed with FASD (30, 69). One study found no differences between children diagnosed with FASD and peers as rated by teachers on the externalising scale of the CBCL (73).

Executive functioning outcomes were reported in four studies comparing children with PAE to non-exposed peers. Children with PAE aged seven to 13 performed more poorly than non-exposed peers on working memory task (digit span) in one study (48) and on a measure of problem solving in another study (50). One study stated poorer category fluency but not letter fluency among children aged seven to 13 with PAE compared to non-exposed peers (53). Mixed results were reported for set-shifting measures in children with PAE compared to non-exposed peers. Children with PAE performed lower than unexposed peers in one study using a Trail-Making Test (53). Lastly, children in the first to third grades with PAE displayed a greater level of teacher-rated hyperactive and impulsive behaviour, compared to non-exposed peers in one study (55).

Executive functioning outcomes were reported in 13 cohort studies comparing varying categories of PAE. One longitudinal study tested executive functioning directly. Children aged 11 to 13 years old with PAE were found to have lower scores on letter fluency compared to non-exposed peers. No differences were reported on category fluency or the WCST (83). Among longitudinal studies measuring executive functioning using proxy-reported measures of executive function, no association was reported between weekly alcohol consumption, or binge drinking in pregnancy and parent-rated executive functioning on the BRIEF at age seven (85). Among the ALSPAC cohort, exposure to binge patterns of PAE (62, 99) but not low levels of PAE (64, 100) was associated with higher hyperactivity and inattention levels, relative to non-exposed peers. Further in the DNBC, there was no association between PAE and SDQ Externalising scores at age five (101) but at age seven children prenatally exposed to alcohol had higher SDQ externalising scores (76, 77). Lastly, in the Oslo Pregnancy Cohort, binge patterns of PAE in early pregnancy were

associated with higher SDQ hyperactivity and inattention scores at age five to six. (74). In a US study, any alcohol exposure was associated with increased odds (OR=1.8 (95% CI=1.0-3.0) of scoring in the clinical range on the CBCL externalising subscale among children aged six to seven years old (102). One study conducted in Australia found no association between alcohol exposure and odds of scoring in the clinical range on the CBCL externalising subscale at low, moderate or high levels of alcohol consumption (75).

Four cohort studies exploring the relationship between average level of PAE and executive functioning outcomes were identified. Average alcohol exposure per day during pregnancy was negatively correlated with measures of working memory and category fluency, but not shift or impulsivity (87). Similarly, a negative association between PAE, and category fluency was found (103). In contrast, no relationship was found between executive functioning assessed using the WCST and the Trail-Making test and average alcohol exposure per day throughout pregnancy (91). Lastly, D'onofrio and colleagues (89) found alcohol exposure was positively associated with conduct problems as measured by the CBCL.

Language

Language includes expressive language skills as expressed by use of language to convey meaning to others. It also includes receptive language skills as expressed by comprehension of language. Twenty studies reported language outcomes.

Seventeen studies compared language outcomes among children diagnosed with FASD to non-exposed peers (33-36, 39, 41-44, 57, 59, 69, 94, 95, 97, 104, 105). Twelve studies examined language outcomes among first-grade students (~7 years old) diagnosed with FASD as part of ACA studies in South Africa, and Italy. Of these twelve studies, eight studies found that children diagnosed with FASD consistently performed more poorly on the Test for Reception of Grammar (TROG) (34, 35, 39, 41-44, 94), and in two studies using the TROG's Italian-language counterpart (57, 59). Lastly, mixed results were observed between two South African studies measuring language outcomes using the Griffiths Mental Development Scale (GMDS) language subscale, a broadband language measure assessing both expressive and receptive language. Adnams and colleagues (33) reported poorer performance than peers amongst children six to eight years old diagnosed with FASD, after adjusting for maternal education, whereas Davies and colleagues (36) failed to replicate these results among children at age five.

A further three studies examined receptive language outcomes amongst children with FASD recruited from specialised FASD diagnostic clinics. In two studies, children with FASD had lower scores than non-exposed peers on the Peabody Picture Vocabulary Test (95, 97), and on the Receptive Language subscale of the Comprehensive Receptive and Expressive Vocabulary Test (CREVT) (105). Further, after controlling for IQ, children with FASD did not differ from non-exposed peers on the Test of Language Competence Figurative Language subscale (69). Lastly, Expressive language skills were reported in two studies, both of which found children with FASD to have poorer expressive language skills than non-exposed peers (104, 105).

One cross-sectional study compared children with categorised PAE to non-exposed peers. Children with PAE were reported to have poorer performance on expressive and receptive language measures (51). In a follow up analysis of a sub-sample matched for IQ, children with PAE did not differ significantly from non-exposed peers. Two longitudinal studies compared language outcomes across varying levels of alcohol exposure. No difference was found between children with PAE and non-exposed peers on the verbal subtest of the McCarthy scales at ages four to five (66). In contrast, one study found children between five and nine who were prenatally exposed to alcohol were reported to have receptive and expressive language difficulties (84).

Memory

Memory is the ability to take in information, store it and recall it later. Assessments of memory included free and cued recall of verbal and visual information, and prospective memory (e.g., the ability to remember and to execute delayed intentions). While there were 10 studies identified in the review which examined working memory, these studies are included in the Executive Function domain, in line with contemporary definitions of executive functioning. In addition to the 10 studies of working memory, an additional 11 studies reported other memory outcomes including free and cued recall, story memory, and prospective memory.

Five studies compared children diagnosed with FASD to non-exposed peers on memory outcomes (37, 94, 97, 98, 106). Children diagnosed with FASD had more difficulties on the three and 30 minute recall condition of the Rey Complex Figure Test (98), greater difficulty recalling verbally presented stimuli (97,

106) and performed more poorly on a prospective memory task (37). In contrast, no differences were reported on a story memory task between children diagnosed with FASD and non-exposed peers in a study of first-grade students (mean= 6.08 years old) (94).

One study identified in the review compared children with PAE, recruited from a clinical population, to non-exposed peers on memory outcomes. Children, between the ages of seven and 12 who had been prenatally exposed to alcohol had lower scores on a spatial recognition task than non-exposed peers (48). Two longitudinal studies compared memory outcomes among categorised groups of children with PAE and non-exposed peers. No differences were found between children between the ages of five and nine with PAE and non-exposed peers on verbal memory (word-span, logical learning) or visual memory (84). In contrast, a study of 11-13 year old children found that children with PAE performed more poorly on the three and 30 minute recall condition of the Rey Complex Figure Test compared to non-exposed peers (83). Two birth cohort studies examined the relationship between the amount of alcohol exposure in pregnancy and offspring memory and found a significant inverse relationship between average alcohol exposure per day during pregnancy and memory performance on the Wide-Ranging Assessment of Learning and Memory at age 10 (91) and the California Verbal Learning Test and at 7.5 years (103).

Motor skills

The motor skills domain includes fine motor skills (coordination of small muscle movement, usually in coordination with the eyes), gross motor skills; (skills which require whole body movement and which involve large core stabilising muscles of the body to perform everyday functions), visual-motor integration (the ability to use input from the visual perceptive system to coordinate fine motor skills), and soft-neurological signs (subtle neurological abnormalities comprising deficits in sensory integration, motor coordination, and sequencing of complex motor acts). Seventeen studies included in the review reported motor skills outcomes.

Twelve studies compared children diagnosed with FASD to non-exposed peers (32, 33, 36, 45-47, 58, 70, 94, 107-109); of these, five studies reported different motor outcomes from the same sample (45-47, 108, 109). Across studies, children diagnosed with FASD were found to have lower composite motor skills (58, 107), fine motor skills (47, 107), and gross motor skills (defined as ball skills) (107) compared to

non-exposed peers. Mixed results were noted on gross motor skills assessments such as balance and strength skills with three studies reporting children diagnosed with FASD to have poorer gross motor skills among children aged five to ten years old (36, 107, 108), while one study found no differences between children with FASD aged between six and eight years old on the GMDS Motor skills (33).

Visual motor integration (VMI) was assessed in five studies of children between five and nine years old, with mixed results. Three studies (32, 70, 94) found children diagnosed with FASD to have poorer VMI compared to non-exposed peers, whereas two studies reported no differences between children with FASD and non-exposed peers on the VMI total scores (46) or Visuo-spatial processing index (58). While Doney and colleagues (46) report no differences on the VMI total score, this is the only study to report the VMI subscales of motor coordination and visual perception, finding children diagnosed with FASD to have greater difficulties in the motor coordination subscale, but not the visual perception subscale. Motor coordination outcomes are reported in a further two studies; one reporting the Developmental Neuropsychological Assessment (NEPSY), Sensorimotor tests (58) and on the Clinical Observation of Motor and Postural Skills (107), both finding children diagnosed with FASD having greater difficulties compared to peers. Lastly, compared to peers, children diagnosed with FASD had greater rates of soft-neurological signs, indicating difficulties in sensory integration, motor coordination, and sequencing of complex motor acts (58, 109).

Five longitudinal studies reported motor skills outcomes among categories of children exposed to alcohol prenatally. No difference was found between five year old children exposed to low (1-4 drinks/wk.), moderate (5-8 drinks/wk.) or heavy (9+ drinks/wk.) levels of alcohol consumption and non-exposed peers on the Movement Assessment Battery for Children in one study (110). Similarly, four to nine year old children with a high level of PAE displayed lower performance on a visual-motor production composite (84). Several studies found no differences on motor skills outcomes between children exposed to alcohol and non-exposed peers. No differences were reported on a manual praxis composite, consisting of the NEPSY sensory-motor function subtest (84), or on the McCarthy scales motor subtest (66). Interestingly, one study found that compared to non-exposed peers, moderate alcohol exposure (3-7 drinks/wk.) was associated with improved motor skills (111). Finally, one study examined the relationship between alcohol exposure

and motor skills outcomes. There was no correlation between average alcohol exposure throughout pregnancy and scores on the grooved pegboard activity (91).

Social skills and social cognition

Socials skills includes skills needed to interact socially and get along with other people, including expressing affection, having friends, showing and recognising emotions, assisting others, using manners, as well as responsibility, rule-following, obeying laws, avoiding victimisation, self-esteem and interpersonal skills (112). Social cognition includes theory of mind: the ability to attribute mental states- beliefs, intents, desires, pretending- to oneself and to others and to understand that these mental states may be different from one's own i.e., false belief tasks, understanding and recognition of emotions. Assessments of social skills, empathy, and social cognition (e.g.,) are discussed below. While there is overlap between social skills and adaptive behaviour, for the purposes of this review, social skills and social cognition are grouped together under a separate domain. A total of 13 studies reported social skills and social cognition outcomes.

Ten studies examined social skills outcomes for children diagnosed with FASD compared to non-exposed peers (30, 38, 58, 72, 95, 113-117). Parent- and teacher-rated social skills were reported in four studies (30, 58, 72, 117). Children diagnosed with FASD aged six to 13 were rated by caregivers as having poorer social skills compared to non-exposed peers in three studies (30, 72, 117). Of the two studies reporting teacher-rated social skills, one study reported poorer social skills among children aged six to 13 years old (30), whereas no difference was reported between children diagnosed with FASD and non-exposed peers aged five to eight years old (58). Children diagnosed with FASD were consistently rated as having greater behavioural problems in social settings compared to non-exposed peers by both caregivers (30, 72) and teachers (30, 58). Children diagnosed with FASD were also rated by caregivers as having lower levels of empathy compared to non-exposed peers (72).

Emotion recognition and discrimination were assessed in three studies comparing children diagnosed with FASD to non-exposed peers (30, 38, 116). Among children aged eight to 12, children diagnosed with FASD differed from non-exposed peers on the ability to correctly identify a target emotion from a picture of the eyes alone (38, 116) and the ability to correctly match photographs of faces to a verbally presented target emotion (30). Children diagnosed with FASD did not differ from peers on the

ability to identify a visually-presented target emotion amongst photographs of full faces (30, 38), or on correctly identifying if the prosody (e.g., the patterns of stress and intonation in language) and content of recorded speech samples were congruent (30).

Four studies comparing children diagnosed with FASD to non-exposed peers assessed theory of mind (30, 38, 72, 95). False belief tasks were reported in four studies. Children aged eight to 12 diagnosed with FASD had more difficulty than peers passing false belief tasks in one study (72) while three studies noted no differences on false-belief tasks across a range of ages from four to eight, six to 13, and nine to 11 (30, 38, 95). Children diagnosed with FASD demonstrated a poorer understanding of how emotion related to social context and to recognise the appropriate affect given various social contexts relative to non-exposed peers in two studies (30, 72). One study reported no differences in the ability to recognise the appropriate emotion for a social situation, to comprehend vignettes describing social interactions including jokes, white lies, sarcasm, or to detect and comprehend a faux pas (38).

Three studies compared children with PAE to non-exposed peers on social skills outcomes (50, 52, 56). Children with PAE were rated as having poorer social skills than non-exposed peers by caregivers, but not teachers (52). Further, there were no differences on either parent or teacher ratings for problem behaviours in social settings (52). Children with PAE displayed greater difficulties than peers in identifying effective responses in social situations, attribution of intent, and in evaluating the outcomes of their actions (50). Lastly, children with PAE were rated as having overall lower playfulness scores than peers (56).

Sleep

Sleep-related behaviours were reported in three studies identified in the review, and one study reported total sleep duration. Sleep and sleep-related behaviours were compared between children diagnosed with FASD and non-exposed peers in two studies (118, 119). Compared with peers, children diagnosed with FASD had greater caregiver reported difficulties in sleep onset delay, daytime sleepiness, bedtime resistance, sleep duration, sleep anxiety, night awakenings and parasomnias, as well as total sleep concerns scores. No differences were noted on sleep disordered breathing in either study. Children with FASD were reported as getting fewer total hours of sleep compared to non-exposed peers (118). Differences in sleep related behaviours were noted among children aged three to six (119), and among a

broader sample of four to twelve year old children(118). In the one longitudinal study to investigate categorised PAE as a risk factor for poor sleep outcomes, at age eight, children prenatally exposed to alcohol had a 2.9-fold increased risk of having short sleep, and 3.6-fold risk of having low sleep efficiency compared to unexposed children (120).

Sensory processing

Sensory processing is defined as the behavioural and functional manifestations commonly associated with sensory processing disorders and includes hypo-, or hyper-, sensitivity to stimulation, poor behavioural regulation, difficulty planning and carrying out motor activities (clumsiness) and emotional reactivity. Three studies compared children diagnosed with FASD to non-exposed peers on sensory processing outcomes (58, 119, 121). Compared to peers, children diagnosed with FASD aged between three and eight years old were reported to have greater sensory processing difficulties as reported by caregivers in two studies, after adjusting for IQ (58), and age (119) respectively. Further, a high proportion of older children with FASD, aged five to 11 years old were identified as having sensory processing issues defined as probable or definite differences on both the short sensory profile (90.9%) and Sensory Processing Measure (81.8%), compared to non-exposed peers, of whom 0% scored in the probably or definite differences range. However, no inferential statistical testing was conducted in this study (121).

Discussion

The aim of this review was to identify and synthesise findings pertaining to cognitive and behavioural functioning among children aged three to 13 years old who have been prenatally exposed to alcohol. We included studies of children with and without a FASD diagnosis, where outcomes had been compared with those of non-exposed and TD children. Using a systematic search procedure, a total of 94 studies examining outcomes associated with FASD or PAE among the target age range were identified. Studies were split into four methodological categories and outcomes grouped into 11 domains of functioning: academic achievement, adaptive behaviour, affect regulation, attention, executive functions, language, memory, motor skills, social skills and social cognition, sleep and sensory processing. Children with FASD and PAE were consistently reported to have differences in observer reports of attention, working memory, hyperactivity and impulsivity, language, and fine motor skills. Lower scores in academic

achievement and adaptive behaviour were also commonly reported. Although examined in fewer studies overall, difficulties with sleep and sleep-related behaviours, and sensory processing were consistently noted in children diagnosed with FASD. While deficits in sensory processing and sleep are considered non-specific for FASD, it is important that assessments be conducted nonetheless as they can contribute valuable information to understanding the difficulties experienced by a child living with FASD for intervention planning (12). Several studies noted that a high proportion of children diagnosed with FASD, and those heavily exposed to alcohol prenatally, were no longer in the care of their birth parents.

This review sought to examine cognitive and behavioural functioning differences that may, or may not, emerge in early childhood and are detectable in preschool and primary school aged children to enable early screening, detection, and intervention. Across the range of the domains identified, differences were identified from a young age on the domains of academic achievement, adaptive behaviour, attention, executive functioning, language, motor skills, sleep, and sensory processing. For example, studies using ACA methodologies recruited first grade cohorts and consistently found differences in hyperactivity and impulsivity, language, and, academic achievement (40, 57, 59). Further, a number of studies highlighted impairments in executive functioning (60, 95, 96), sleep and sensory processing (119) and early maths skills in samples including children in preschool. This review highlights that children with FASD are experiencing difficulties in multiple domains of cognitive and behavioural functioning through preschool and into the early years of schooling. Without identification and provision of appropriate supports, these early difficulties are likely to increase and compound as children develop and the demands placed upon children increase.

Methodological considerations

In an attempt to synthesise the studies included in this literature review and the breadth of methods used to examine cognitive and behavioural impacts of PAE, the studies were divided into four broad methodological categories: (1) Studies comparing cognitive behavioural outcomes in children diagnosed with FASD to non-exposed peers, (2) Studies comparing children with PAE and no alcohol exposure, defined based on retrospective report of PAE, recruited where concerns about behaviour existed, or from populations believed to have a high level of alcohol exposure, (3) Longitudinal studies of children recruited

during pregnancy which compared subsequent cognitive and behavioural outcomes examined across varying categorised PAE levels (i.e., across low, moderate, and heavy groups), and (4) Longitudinal studies of children recruited during pregnancy examining the relationship, or lack thereof, between PAE, measured as a continuous variable (i.e., average number of drinks per day during pregnancy) and later outcomes. However, there was considerable methodological variation across studies that were grouped together, as discussed above, which made the research challenging to compare and highlights the complexity of prenatal alcohol exposure and outcomes.

While children diagnosed with FASD and/or PAE broadly showed greater cognitive and behavioral deficits across identified studies, there are several factors that may have impacted study outcomes that should be taken into consideration when interpreting the findings of these studies, as well as when planning future research. These include familial characteristics of children with FASD or PAE, the age of assessment and emergence of cognitive and behavioural outcomes, the measurement of cognitive and behavioural outcomes, and the measurement of alcohol exposure.

A sizeable proportion of studies examining the cognitive and behavioural functioning of children with FASD identified in this review were conducted in South Africa. Studies from South Africa accounted for 12 of the 50 (24%) studies comparing children with FASD to non-exposed peers, and two of the nine studies comparing children with PAE to non-exposed peers (see Table 2). These studies were primarily ACA studies in disadvantaged regions aimed at determining FASD prevalence.

Family characteristics of children living with FASD

A trend was observed among the studies of children with FASD, and of children heavily exposed to alcohol prenatally for a substantial proportion of the clinical sample (e.g., children with FASD or PAE) being reported to be in the care of caregivers other than their birth parents. Among the studies of children with FASD, 27 of 50 studies reported this information with the proportion of children in care other than their birth parents ranging between 32% and 100%. Further, among the nine studies comparing groups of children with PAE to peers, a similarly high proportion of children no longer in the care of their birth parents was reported; 26.7% to 95%. It is important to note that studies conducted in South Africa represent the lower figures among both groups of studies which may reflect differences in social services across

countries. This bias may reflect the populations that seek assessments from clinics that supported recruitment efforts in most studies, given that in many studies populations of children with FASD were recruited through behavioural clinics. Importantly, while several studies identified used ACA methodologies in randomly selected schools to identify FASD prevalence estimates in the US, Italy, and South Africa, only one South African study reported the proportions of children who were no longer in the care of their parents.

This overlap with the population of children in out of home care where removal from birth families and placement into other types of care may have been for PAE but may also have been for other in utero exposures (e.g., illicit drugs) and/or subsequent abuse and/or neglect in childhood. Experiences of early life adversity are associated with changes in brain structure (122), and adverse cognitive and behavioural functioning outcomes in children including difficulties in emotional and behaviour regulation, executive functioning, and interpersonal difficulties (123, 124). Exposure to abuse and neglect experiences are potentially a contributing factor for cognitive and behavioural presentations of children with FASD; for example, abuse and trauma related symptomatology in addition to those associated with brain development impairments associated with in utero alcohol exposure. Research has recently begun to examine in greater detail the interaction between experiences of maltreatment and PAE (125).

The longitudinal birth cohort studies varied in how early life adversity was addressed. In several studies, this information was not reported (e.g., the ALSPAC studies), or children prenatally exposed to alcohol who were removed from the care of their birth parents were excluded at follow up. Two cohort studies; the DNBC and the UK Millennium study included a quality of home environment variable[7] as a co-variate in the analysis (65, 76, 77, 85, 86, 101, 110, 126). While there do appear to be some consistent behavioural and cognitive functioning differences emerging between children with FASD and peers, the

[7] The Danish National Birth Cohort, and Lifestyle During Pregnancy Study included a Family/Home Environment variable, defined as the sum of the following indicators; living with only one biological parent; changes in primary care givers; daycare for more than eight hours/day before age three; ≥14 days of separation from parents; breakfast irregularities; maternal depression and maternal/paternal alcohol intake above the official recommendations; an index of the child's health status.

The Millennium Cohort defined family context as: number of children in the household; child's age; highest parental educational qualification; parental income; mother's mental health; parental discipline strategies; mother's self-rated competence; mother's self-rated closeness of relationship with child; whether the mother currently drank alcohol; frequency of someone reading to the child; whether the child had regular bedtimes.

high proportion of children who are no longer reported to be in the care of their birth parents confounds these findings. The overlap with an out of home care population where removal/placement of children may have been for in utero alcohol exposure but may also have been for other in utero exposures (e.g., illicit drugs) and/or subsequent abuse and/or neglect in childhood – hence comprehensive assessment must be undertaken to determine aetiology of differences.

Age of assessment, and emergence of cognitive and behavioural outcomes

The age at assessment is an important consideration when observing behaviours that show an age-related emergence. There are rapid changes across the domains of cognitive and behavioural functioning identified in this review. Further, most younger children demonstrate difficulties with self-regulation and fine motor skills. Difficulties might be expected to emerge later; for example, children with FASD may not develop skills in executive functioning at the same rate as their peers, and differences between children with FASD and their peers may widen as children age.

Importantly difficulties in some domains were noted from pre-school, including early maths difficulties (60), adaptive behaviour (68), executive functioning (e.g., working memory, set-shifting, impulsivity/inhibition (60, 95, 96), language difficulties (51, 95), sleep, and sensory processing (119). Among slightly older children (those in the first grade, and approximately 6-7 years old), numerous ACA studies identified difficulties in working memory, language, and proxy reports of attention (39-44, 57, 59, 70). While language skills were assessed among first grade children in twelve ACA studies, these were all conducted in South Africa (k = 10) and Italy (k = 2). In contrast, studies examining language outcomes between children with FASD, or those heavily exposed to alcohol, and non-exposed peers conducted in Canada and the US were typically examined among older children (i.e., 6 to 13, 8 to 12, 9 to 11).

The age at assessment may mask also some potential difference. For example, several studies investigating theory of mind and false belief tasks were identified and no differences between children with FASD and non-exposed peers were reported among studies of children aged between four and eight (95), six to 13 (30) and nine to 11 (38). Children can reliably pass false-belief tasks by age four and these studies included children approximately seven years of age and showed a ceiling effect (i.e., most children across groups got the answers correct). A question remains about whether the children with FASD developed

these skills at the same age as peers, or whether the development was delayed, albeit before participation in the study.

Measurement of cognitive and behavioural outcomes

In the current review, contrasting results were seen between direct testing of children and observer reports of the same domain. This was particularly noted in the attention domain, reflecting the gap wherein children with FASD present with behavioural symptoms of inattention, but do not always demonstrate deficits on direct assessments (32, 82). These findings may be explained, in part, by the exclusion of children who were unable to complete direct assessment tasks. In these studies, a high proportion of children with FASD were unable to successfully complete attention tasks. Contrasts between cognitive and behavioural functioning as measured by direct assessment (i.e., clinical or experimental measures) and observer-reports of behaviour should be a focus of future studies. Both methods of assessment, however, have limitations. Direct assessments have been criticised for lacking ecological validity and giving an inaccurate impression of real-world functioning as they require children to sit in a distraction free room, one-on-one with the tester who may be encouraging children to stay focused. On the other hand, observer reports have been criticised as being prone to bias and not reflecting objective measures (127). Future research should explore novel methods for ensuring ecological validity (128) and ensure that information is collected from multiple sources (e.g., direct assessment, parent report, and teacher report).

Measurement of alcohol exposure

Binge drinking was consistently associated with poorer outcomes in the domains of academic achievement (63), affect regulation (74, 76), hyperactivity, and executive function (62, 77). Similarly, heavy alcohol use in longitudinal studies showed the clear associations with adverse cognitive and behaviour function outcomes at follow up (75, 86). In contrast, other categories of alcohol use (as opposed to misuse) show fewer clear patterns. Previous research has highlighted the deleterious impact of binge drinking during pregnancy on child cognitive and behavioural outcomes (129). Examining functioning difficulties across alcohol exposure levels was confounded by country-specific variation in the definition of a standard drink, and the level of alcohol consumption that is defined as binge drinking. For example, a standard drink is defined as 8gm alcohol in the ALSPAC studies conducted in the UK (62, 64, 99, 100), as 10gm in Australian

cohort studies (61), and as 12gm in DNBC studies (76, 77, 85). Definitions of binge drinking varied between four drinks or more per occasions in the ALSPAC in the UK (62) and the RASCALS study in Australia (61), and five drinks or more per occasion in the DNBC studies in Denmark (77). Results were further confounded by the frequency of binge drinking. Several studies used a dichotomous exposure to at least one episode of binge drinking throughout the whole pregnancy, whereas others used a count of the binge drinking episodes throughout pregnancy (e.g., 0, 1, 2, 3 or more), and some studies narrowed the definition to specific windows (e.g., episodes of binge drinking in the first six weeks of pregnancy, or contrasting reports of binge drinking in the first or third trimesters only). Interestingly, several studies suggested a protective effect of alcohol; suggesting that low levels of alcohol consumption were associated with *improved* cognitive and behavioural functioning. It is suggested however, that this may be due to the relationships between alcohol consumption and variables such as higher maternal education and income (76, 111).

Studies frequently reported that the comparison groups of TD peers were screened for alcohol exposure before study enrolment but relied heavily on retrospective alcohol exposure reports. There is considerable debate around the accuracy of retrospective reports, owing both to the length of time at follow up, and the sensitive nature of the topic (130). Further, several studies identified they were unable to directly collect this information from control groups over concerns of the sensitive nature, and relied instead on participants to volunteer this information and opt out of the study, if they reported alcohol exposure (80, 81, 107). Collection of alcohol exposure information is further complicated by the high proportion of children across studies who were identified as no longer being in the care of their birth parents. In these studies, alcohol exposure was determined by review of collateral reports including medical, legal, or social services information, or by informant-report. Identification of PAE retrospectively is a challenge for the field broadly, and these studies reflect the need for accurate routine screening for PAE in health care settings.

FASD Diagnostic sub-groups

Outcomes across the FASD diagnostic subgroups (i.e., FAS vs pFAS vs ARND) were compared in seven studies (38-44). FAS and pFAS groups were compared in seven studies, with ARND included in three of these seven studies. Six studies reported all clinical groups to differ from non-exposed peers (38, 39, 41-44). The seventh study found those with pFAS, but not FAS to differ from non-exposed peers on

most outcomes included in the study (40). While a small number of studies found differences between the FASD diagnostic subgroups, the most consistent result was no differences between sub-groups. At the outset of data analysis, several studies presented a decision point wherein children across diagnostic subgroups were compared and a decision to treat as one group or split into various categories was made. A recommendation for future studies is to either undertake this, or alternatively present a sensitivity analysis where FASD subgroups are pooled and compared to peers.

In addition to the studies comparing diagnostic subgroups, eight studies comparing children with FASD to non-exposed peers also included a comparison group of children with PAE but not diagnosed with FASD (32, 36-38, 40, 41, 44-47). Children with PAE but not diagnosed with FASD did not differ significantly from either FASD cases or non-exposed comparison cases on measures of working memory, language, attention (41), or motor skills (45, 47) whereas the heavy PAE group performed better than those diagnosed with FASD on social cognition (38), visual motor integration (46), and prospective memory (37). The PAE group did not differ from unexposed peers. Lastly, one study, although including both a FASD and a heavy PAE group but not diagnosed with FASD, did not compare outcomes of these two groups (36).

Strengths and limitations of this review

In line with scoping review methodology (23, 24), no quality assessment was conducted for included studies. Without assessing the methodological quality, there may some some risk of bias in the studies included in the literature review. Further, a lack of quality assessment can make scoping reviews more difficult to interpret (24). Secondly, a number of potentially relevant studies were identified in the database searches but were excluded as the sample bridged the age range for inclusion (i.e., the mean age may have fallen within the included age range, but included children from six to 18). The aim of this study was to examine the cognitive and behavioural outcomes associated with prenatal alcohol exposure among early school-aged children with FASD. The decision to exclude studies outside the age range of interest was made in recognition of the rapid cognitive development through childhood, and age-related differences noted among children with FASD. Given that this study sought to examine the cognitive and behavioural profiles to support opportunities for earlier screening, identification and intervention, studies which fell outside the target age range were excluded. Previous research has shown age-related changes

in cognitive and behavioural domains for childen with FASD. For example, Taylor and Enns (131) reported that compared to school-aged (six to 12 years old) children with FASD, adolescents (12 to 18 years old) with FASD, showed lower overall IQ scores, poorer verbal comprehension skills and arithmetic compared to age-related norms. Similarly, Tamana and colleagues (132) reported that older children with FASD showed greater deficitis in executive functioning and processing complex information.

Implications

This review highlights a broad spectrum of impairments associated with PAE. While the cognitive and behavioural presentation of every child is unique, there do, however, appear to be some consistent findings across studies about domains in which children with FASD/PAE may demonstrate different performance compared to peers including academic achievement, adaptive behaviour, observer reports of attention, working memory, hyperactivity and impulsivity, language, and fine motor skills. The results of this review suggest that when children present with concerns in any of these domains, PAE should be assessed as part of developmental history taking. Developmental history taking is especially important, albeit more complicated, if the child is no longer in the care of their birth parents. This review highlights that cognitive and behavioural functioning difficulties may be apparent from a young age, including difficulties with education from the first grade. The need for early intervention to support children prenatally exposed to alcohol, including those meeting FASD diagnostic criteria is clearly evident.

These results have important implications for the early assessment of and intervention for children prenatally exposed to alcohol. The most recent systematic review of interventions for children with FASD reports promising results for interventions aimed at increasing children's self-regulation, with improvements noted in parent/caregiver reports of behaviours and neuropsychological testing of executive functioning (133). In addition to specific interventions, the early identification of FASD is associated with better long-term outcomes (16). Early identification enables families to be equipped with the knowledge and strategies that have been shown to be effective in managing their children's behaviour and minimise confounding of the factors associated with poor long-term outcomes. This review suggests that children with FASD, and prenatally exposed to alcohol demonstrate observable cognitive and behavioural difficulties during the preschool years, and as they transition to school. It is vital that children who may be experiencing these

cognitive and behavioural difficulties are identified early, and opportunities given for intervention to minimise the risk for adverse long term outcomes associated with FASD. In addition, transition periods in the school years may present opportunities for on-going screening and assessment of children's needs (134).

Conclusions

This review examined cognitive and behavioural functioning associated with diagnosed FASD and PAE amongst children aged between three and 13 years old. PAE in general was associated with poorer performance in a range of cognitive and behavioural domains, but most consistent were differences in working memory, hyperactivity/impulsivity, language, fine motor skills, and observer reports of inattention. Further, difficulties in academic achievement and adaptive behaviour were also commonly reported. Methodological variation between studies that were grouped together, and across the groups limits clear trends that can be drawn from the literature; such as the high proportions of children who are no longer in the care of their birth parents, and the potential overlap with experiences of abuse and neglect which may impact upon cognitive and behaviour functioning of children with FASD and PAE. This information will contribute to early identification and intervention for children with PAE/FASD to improve long term outcomes.

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Supporting Information[8]

S1 Table. Characteristics of studies comparing children with FASD to Non-exposed peers

S2 Table. Characteristics of studies comparing children with PAE to Non-exposed peers

S3 Table. Characteristics of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol

S4 Table. Characteristics of studies examining alcohol exposure as a continuous variable

[8] Supporting tables are presented in Appendix O.

Tables

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
Academic Achievement *K*= 7, *n*= 683	IDPA	Aragon et al., (2008)	Italy	80	6.13(0.34)	6-7	FASD < CON, p = 0.004	Among the seven studies reporting academic outcomes, three studies reported academic achievement (Aragón et al., 2008; Coles et al., 1997; Kodituwakku, Coriale, et al., 2006), and all found children with FASD to have lower achievement compared to non-exposed peers. Four studies report arithmetic outcomes (Adnams et al., 2001; Jirikowic, Olson, & Kartin, 2008; May et al., 2015; Rasmussen & Bisanz, 2011), with all studies reporting children with FASD performing more poorly on arithmetic tasks compared to non-exposed peers. Two studies reported literacy outcomes (Jirikowic, Olson, et al., 2008; May et al., 2015). Both studies found children with FASD to have lower spelling performance but contrasting results on reading performance.
		Koditukwakku et al., (2006)	Italy	82	6.3(0.46)	6-7	FASD < CON, p < 0.001	
	K-ABC Achievement composite score	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	FAS/FAE < CON, p < 0.02	
	WRAT-R	Jirikowic, Olson et al., (2008)	USA	51	6.5(0.88)	5-8	Spelling; FASD<CON, p=0.03 Reading; FASD=CON, p=0.37 Arithmetic; FASD<CON, p<0.001 Adj. for IQ	
		May et al., (2015)	USA	216	7.45(NR)	NR	Spelling; FASD<CON, p<0.001 Reading; FASD<CON, p<0.001 Arithmetic; FASD<CON, p<0.001	
	GDMS Practical Reasoning	Adnams et al., (2001)	South Africa	68	7.12(0.5)	NR	FAS<CON, p<0.001	
	WJ-III Maths subscale	Rasmussen & Bisanz (2011)	Canada	41	5.67(NR)	4-6	FASD<CON, p<0.01	
Adaptive Behaviour *K*= 7, *n*= 763	Vineland Adaptive Behaviour Scales (VABS)	May et al., (2014)	USA	216	6.96(0.54)	NR	Total Scores; FASD<CON, p<0.001	Children with FASD performed more poorly than their peers on the observer-reported VABS and SIB-R in five studies (May et al., 2014; May et al., 2015; Quattlebaum & O'Connor, 2013; Thomas et al., 1998). Impairments were noted in the sub-scales measuring language and communication, social skills, and daily living skills regardless of the measure used. Parent reported motor skills were not different between children with FASD and typically developing peers (Jirikowic, Kartin, & Olson, 2008). Contrasting results were reported in two South African studies reporting the clinician-administered GMDS Personal-Social Scale (Adnams et al., 2001; Davies et al., 2017), with one study controlling for maternal education reporting no differences between children with FASD and peers. Children with
		May et al., (2015)	USA	212	7.13(0.48)	NR	Total Scores; FASD<CON, p<0.003 Communication; FASD<CON, p<0.001 Socialisation; FASD<CON, p<0.003 Daily living; FASD<CON, p<0.001	
		Quattlebaum & O'Connor (2013)	USA	125	8.18(2.10)	6-12	Communication; FASD<CON, p<0.001, d=1.38 Socialisation; FASD<CON, p<0.001, d=0.88 Daily living skills; FASD<CON, p<0.001, d= 1.34 Adj. for IQ	
		Thomas et al., (1998)	USA	45	10.3(2.0)	5-13	Socialisation; FASD<CON, p<0.05 Adj. for SES	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
	Scales of Independent Behaviour-Revised (SIB-R)	Jirikowic, Kartin et al., (2008)	USA	48	6.5(0.88)	5-8	Social/communication; FASD<CON, p=0.007 Personal living; FASD<CON, p=0.003 Community living; FASD<CON, p=0.017 Motor skills; FASD=CON, p=0.229 Adj. for IQ	FASDwere reported to perform more poorly than peers in a larger, subsequent study noting that maternal education was not included as a covariate in this study.
	GMDS Personal-Social subscale	Adnams et al., (2001)	South Africa	68	6.99(0.5)	6.3-8	FAS=CON, p NR, Adj. for maternal IQ	
		Davies et al., (2017)	South Africa	121	NR	5-6	FAS<CON, p<0.001	
Affect Regulation *K*= 5, *n*= 535	Child Behaviour Checklist (CBCL) Internalising Scale	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	Parent/Caregiver; FAS/FAE=CON, p NR	Among the five studies reporting parent or caregiver rated internalising behaviour from the CBCL, two studies reported children with FASD to display greater internalising behaviour than typically developing peers (Quattlebaum & O'Connor, 2013; Stevens et al., 2015), whereas three studies reported no differences (Coles et al., 1997; Greenbaum et al., 2009; Tsang et al., 2017). Among the subset of papers reporting teacher-rated internalising behaviour, children with FASD were consistently rated as displaying greater internalising behaviours compared to their typically developing peers (Greenbaum et al., 2009; Quattlebaum & O'Connor, 2013; Tsang et al., 2017). Lastly, children with FASD were reported to have a higher level of depressive symptoms in one study, after adjusting for differences in IQ (Quattlebaum & O'Connor, 2013).
		Greenbaum et al., (2009)	Canada	97	9.23 (NR)	6-13	Parent/caregiver; FASD=CON, p NR Teacher: FASD>CON, p<0.001 Adj. for Abuse history, SES, IQ	
		Quattlebaum & O'Connor (2013)	USA	125	8.18(2.10)	6-12	Parent/caregiver; FASD>CON, p<0.001, d=1.06 Teacher; FASD>CON, p<0.05, d=0.58 Adj. for IQ	
		Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	Parent/caregiver; FASD>CON, p<0.001, η^2= 0.35 Adj. for gender	
		Tsang et al., (2017)	Australia	108	8.7(NR)	7-9	Parent/caregiver; FASD=CON, p NR Teacher; FASD>CON, p=0.02	
	Pictorial Depression Scale (PDS)	Quattlebaum & O'Connor (2013)	USA	125	8.18(2.10)	6-12	FASD>CON, p<0.05, d=0.70 Adj. for IQ	
Attention *K*=11, *n*= 2,129	Continuous Performance Task (CPT)	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	FASD not different from non-exposed peers on any CPT indices (p values not reported)	Among the five studies reporting direct assessments of attention, mixed results were reported. Studies noted children with FASD did not differ from peers with regards to their sustained attention (Coles et al., 1997; Kooistra, Crawford, Gibbard, Kaplan, &
		Kooistra, Crawford, Gibbard,	Canada	116	8.83(1.17)	7-10	FASD group was more variable in response (p<0.001) and made more errors of omission than non-	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
		Ramage, & Kaplan (2010)					exposed peers (p=0.001), but not errors of commission (p NR) (Kooistra et al., 2010)	Fan, 2011), nor on assessments of switching attention (Kooistra et al., 2011; Lane et al., 2014). Methodological differences, such as recruiting a control group matched on mental, rather than chronological age (Lane et al., 2014) or failure of participants with FASD to complete a task (Coles et al., 1997) limit conclusions that can be drawn. Eight studies reported indirect measures of attention (i.e., parent or teacher rated). Of these, parents rated children with FASD as having greater attention difficulties in 7 studies (Coles et al., 1997; Kodituwakku, Coriale, et al., 2006; Lane et al., 2014; May, de Vries, et al., 2016; May et al., 2017; May, Marais, et al., 2016; Quattlebaum & O'Connor, 2013). One study reported no differences between children with FASD and non-exposed peers as rated by parents (Aragón et al., 2008). Greater attentional difficulties among indviduals with FASD were reported by teachers in two studies (Aragón et al., 2008; Kodituwakku, Coriale, et al., 2006).
	Vigilance Task	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	FASD not different from non-exposed peers on any CPT indices (p NR)	
	Test of Everyday Attention (TEACH-5)	Lane et al., (2014)	Canada	28	11.73(1.36)	8-13	Selective attention subtests Sky search correct; FASD=CON, p=0.093 Timing per correct target; FASD=CON, p=0.147 Map mission targets found; FASD>CON, p=0.018 Attention control/switching subtests Creature counting correct; FASD<CON, p=0.003 Timing; FASD=CON, p=0.782 Opposite worlds sequential condition; FASD=CON, p=0.804 Opposite words switching condition; FASD=CON, p=0.715	
	Attention Network Task	Kooistra et al., (2011)	Canada	113	8.81(1.25)	7-10	Alerting (Sustained attention); FASD=CON, p NR Orientating (switching attention); FASD=CON, p NR Conflict (error detection and inhibition); FASD>CON, p=0.012, partial η^2=0.107) Adj for Age, SES, IQ, gender	
	Prospective Memory on-going task absorption	Lewis et al., (2016)	South Africa	89	11(0.5)	NR	FASD>CON, (indicating lower performance) p<0.05	
	CBCL Attention problems	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	Caregiver rated; FASD>CON, p<0.001	
	TRF (Teacher)	May, de Vries, et al., (2016)	South Africa	553	7.23(0.80)	NR	FASD>CON, p<0.001	
		May et al., (2017)	South Africa	388	7.21(0.84)	NR	FASD>CON, p<0.001	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
		May, Marais, et al., (2016)	South Africa	406	6.93(0.63)	NR	FASD>CON, p<0.001	
	Swanson, Nolan and Pelman Questionnaire, 4th Ed (SNAP-IV) Attention	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	FAS/FAE>CON, p<0.001	
		Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	FASD>CON, p<0.001, d=1.52 Adj. for IQ	
	DBD Attention	Aragón et al., (2008)	Italy	80	6.13(0.34)	6-7	Parent; FASD=CON, p=0.100 Teacher; FASD<CON, p=0.005	
		Kodituwakku, Coriale, et al., (2006)	Italy	82	6.3(0.46)	6-7	Parent rated; FASD<CON, p=0.013 Teacher; FASD<CON, p<0.001	
	Conner's Rating Scale	Lane et al., (2014)	Canada	28	11.73(1.36)	8-13	FASD>CON, p NR	
Executive Functioning *K*= 21, *n*= 3,040	Executive Function Scale for Early Childhood	Fuglestad et al., (2015)	USA	89	4.4(0.76)	3-5	FASD<CON, p<0.001, η^2= 0.14 Adj. for age.	Within executive functioning, hyperactivity and impulsivity domains, there are several sub-domains, namely fluency, inhibition, planning, set shifting, working memory and hyperactivity/impulsivity. Children with FASD were consistently reported to have impairments in working memory (as assessed using digit span) (Kalberg et al., 2013; May et al., 2013; May, de Vries, et al., 2016; May et al., 2017; May et al., 2015; May, Marais, et al., 2016; Rasmussen, Wyper, & Talwar, 2009) or composite measures (May et al., 2014). A single study reported no differences between children with FASD and non-exposed peers on digit span (Quattlebaum & O'Connor, 2013) In contrast, no differences were reported in two studies using a block span test (Rasmussen & Bisanz, 2011; Rasmussen, Wyper, et al., 2009). Children with FASD were more impaired than peers on measures of set-shifting (Coles et al., 1997; Fuglestad et al., 2015; Quattlebaum & O'Connor, 2013) and inhibition tasks (Fuglestad et al., 2015; Kooistra et al., 2010; Rasmussen, Wyper, et al.,
	Wisconsin Card Sorting Test	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	Categories completed; FAS/FAE CON, p<0.04 No significant differences of number of correct cards or errors, or types of errors (e.g., perseverative errors) (All p NR)	
	Children's Colour Trails	Quattlebaum & O'Connor (2013)	USA	125	8.18(2.10)	6-12	Form 1 Consecutive counting; FASD=CON, p NR, d=0.54 Form 2 Switching by colour; FASD<CON, p<0.05, d=0.7 Adj. for IQ	
	Letter and Category Fluency	Kodituwakku, Adnams, et al., (2006) [1]	South Africa	123	7.63(0.91)	6-9	Letter fluency; FASD<CON, p<0.001, d=0.58 Category fluency; FASD<CON, p<0.001, d=0.43	
	Working Memory composite	May et al., (2014)	USA	216	6.96(0.54)	NR	FASD<CON, p<0.001	
	Digit span	Kalberg et al., (2013)	South Africa	113	6.54(0.63)	NR	FASD<CON, p<0.001	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)
		May et al., (2013)	South Africa	277	7.12(0.73)	NR	FASD<CON, p<0.001
		May, de Vries, et al., (2016)	South Africa	553	7.23(0.80)	NR	FASD<CON, p<0.001
		May et al., (2017)	South Africa	388	7.21(0.84)	NR	FASD<CON, p<0.001
		May et al., (2015)	USA	216	7.13(0.48)	NR	FASD<CON, p<0.001
		May, Marais, et al., (2016)	South Africa	406	6.93(0.63)	NR	FASD<CON, p<0.001
		Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	FASD=CON, p N.S, d=0.50 Adj. for IQ.
		Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	FASD<CON, p<0.05
	WMTB-C	Rasmussen & Bisanz, (2011)	Canada	41	NR	4-6	Digit Recall; FASD<CON, p<0.01 Word-list matching; FASD=CON, p N.S Word-list recall FASD<CON, p<0.01 Nonword list recall FASD<CON, p<0.01 Block recall FASD=CON, p N.S Backward digit recall; FASD=CON, p N.S
		Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	Digit Recall Block Recall; FASD=CON, p NR
	DayNight	Rasmussen, Pei, et al., (2009)	Canada	70	NR	5-10	FASD=CON, p NR
		Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	FASD<CON, p<0.01
	Whispers	Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	FASD<CON, p<0.01

2009). One study reported no differences between children with FASD and non-exposed peers on a measure of inhibition (Rasmussen, Pei, Manji, Loomes, & Andrew, 2009).

A single study reported fluency outcomes, noting children with FASD had lower scores than non-exposed peers (Kodituwakku, Adnams, et al., 2006).

No studies reporting planning tasks (e.g., Tower of Hanoi task) were identified.

On indirect measures of executive function, children with FASD were reported to have greater behaviour regulation difficulties (BRIEF Behavioural Regulation Index), and more difficulty in self-managing and monitoring their own performance (BRIEF Meta-cognition index) (Quattlebaum & O'Connor, 2013).

Parents and teachers generally rated children with FASD as displaying more hyperactive behaviours, and greater levels of impulsivity compared to typically developing peers (Kodituwakku, Coriale, et al., 2006; Quattlebaum & O'Connor, 2013), although one study reported a contrasting result (Aragón et al., 2008).

Children with FASD were also rated as displaying a greater level of externalising behaviour relative to their peers, by both parents (Greenbaum et al., 2009; Quattlebaum & O'Connor, 2013; Stevens et al., 2015) and teachers (Greenbaum et al., 2009; Quattlebaum & O'Connor, 2013). Differences in externalising behaviour were not consistent, and two studies reported no differences between children with FASD and their non-exposed peers (Coles et al., 1997; Tsang et al., 2017).

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
	Rey-Osterrieth Complex Figure Task	Pei, Job, Kully-Martens, & Rasmussen, (2011)	Canada	70	8.29(NR)	6-12	Organisation; FASD>CON, $p<0.01$ Accuracy; FASD>CON, $p<0.001$ Style; FASD=CON $p>0.05$ Error; FASD=CON, $p<0.09$	Null results Quattlebaum & O'Connor 2013- trail making A: doesn't engage the switching element so considerably lower task load. Contributes to a null results. Note similar paper in the PAE studies (Vaurio et al.) In the studies using block span measures, poorer performance by the CON group may be contributing to the null results. (60, 95). For example, Rasmussen, Wyper et al (2009) report the mean score for the CON group to be approximately 70, on a score normed for a mean of 100 and standard deviations of 15. Among the studies reporting the CBCL externalising scale, Coles et al., (1997) and Tsang et al., (2017) are both conducted in disadvantaged setting; Tsang in remote Aboriginal communities and Coles in disadvantaged African American communities, which may contribute to null results, compared to the other studies which recruited the comparison groups through community postings, or were the biological children of the foster or adoptive parents of children with FASD.
	Go-No Go inhibition tasks	Kooistra et al., (2010)	Canada	116	8.83(1.17)	7-10	FASD<CON; standard deviation of response latency $p=0.001$, error percentage $p=0.001$ FASD=CON; response latency $p=0.088$, omission (p N.S), or commission errors (p NR)	
	Delay of Gratification	Fuglestad et al., (2015)	USA	89	4.4(0.76)	3-5	FASD>CON, $p<0.001$, $\eta^2= 0.16$ (minutes waited) Adj. for age	
	BRIEF	Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	Behavioural regulation; Parent rated FASD>CON, $p<0.001$, $d=1.75$ Teacher rated FASD>CON, $p<0.001$, $d=1.43$ Meta-cognition; Parent rated FASD<CON, $p<0.001$, $d=1.82$ Teacher rated FASD<CON, $p<0.05$, $d=0.69$ Adj. for IQ	
	DBD Hyperactivity/ impulsivity subscale	Aragón et al., (2008)	Italy	80	6.13(0.34)	6-7	Teacher; FASD=CON, $p=0.691$ Parent rated: FASD=CON, $p=0.836$	
		Kodituwakku, Coriale, et al., (2006)	Italy	82	6.3(0.46)	6-7	Teacher rated; FASD>CON, $p=0.032$ Parent rated; FASD>CON, $p=0.015$	
	SNAP-IV Hyperactivity/Impulsivity subscale	Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	FASD<CON, $p<0.001$, $d=1.77$ Adj. for IQ	
	CBCL Externalising Behaviour subscale	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	Parent- rated form; FAS/FAE=CON, $p=$ NR	
		Greenbaum et al., (2009)	Canada	97	9.23 (NR)	6-13	Parent; FASD>CON, $p<0.001$ Teacher-Rated Form; FASD>CON, $p<0.001$ Adj. for abuse history, SES, IQ	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
		Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	Parent; FASD>CON, $p<0.001$, $d=2.00$ Teacher; FASD>CON, $p<0.001$, $d=1.24$ Adj. for IQ	
		Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	Parent: FASD>CON, $p<0.001$, $\eta^2=0.61$ Adj. for gender.	
		Tsang et al., (2017)	Australia	108	8.7(NR)	7-9	Parent; FASD=CON, $p>0.05$ Teacher-Rated; FASD=CON, $p=0.07$	
Language and Communication *K*= 17, *n*= 2,986	Test for Reception of Grammar	Kalberg et al., (2013)	South Africa	113	6.54(0.63)	NR	FASD<CON, $p<0.001$ Adj. for age.	Children with FASD performed more poorly than non-exposed peers on measures of receptive grammar including the TROG (Kalberg et al., 2013; Kodituwakku, Adnams, et al., 2006; May et al., 2013; May, de Vries, et al., 2016; May et al., 2017; May et al., 2007; May, Marais, et al., 2016; Viljoen et al., 2005), and the Italian language Rustioni Test of Language Competence (Aragón et al., 2008; Kodituwakku, Coriale, et al., 2006). Three studies reported impairments in receptive vocabulary using the CREVT (Wyper & Rasmussen, 2011) and the PPVT (Rasmussen, Pei, et al., 2009; Rasmussen, Wyper, et al., 2009). Children diagnosed with FASD did differed from non-exposed peers on the Test of Language Competence, however differences were attenuate and no longer significant after controlling for IQ (Quattlebaum & O'Connor, 2013). Controlled for IQ- no other study did so, Test of Language Competence did differ between groups prior to IQ adjustment. Contrasting results were found on the GMDS language subscale among two studies of similar aged children from South Africa (Adnams et al., 2001; Davies et al., 2017).
		Kodituwakku, Adnams, et al. (2006)	South Africa	123	7.63(0.91)	NR	FASD<CON, $p<0.001$	
		May et al., (2013)	South Africa	277	7.12(0.73)	NR	FASD<CON, $p<0.001$	
		May, de Vries, et al., (2016)	South Africa	553	7.23(0.80)	NR	FASD<CON, $p<0.001$	
		May et al., (2017)	South Africa	388	7.21(0.84)	NR	FASD<CON, $p<0.001$	
		May et al., (2007)	South Africa	218	7.81(0.86)	NR	FASD<CON, $p<0.001$	
		May, Marais, et al., (2016)	South Africa	406	6.93(0.63)	NR	FASD<CON, $p<0.001$	
		Viljoen et al., (2005)	South Africa	210	6.54(0.64)	NR	FASD<CON, $p<0.001$	
	Rustioni Test of Language Competence	Aragón et al., (2008)	Italy	80	6.13(0.34)	6-7	FASD<CON, $p=0.028$	
		Kodituwakku, Coriale, et al., (2006)	Italy	82	6.3(0.46)	6-7	FASD<CON, $p<0.001$	
	CREVT	Wyper & Rasmussen, (2011)	Canada	50	9.0(NR)	5-13	Receptive Vocabulary; FASD<CON, $p<0.01$ Expressive Vocabulary; FASD<CON, $p<0.01$	
	PPVT	Rasmussen, Pei, et al., (2009)	Canada	70	NR	5-10	FASD<CON, $p<0.01$	

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Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
		Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	FASD<CON, $p<0.01$	
	Test of Language Competence-Figurative Language	Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	FASD=CON, p NR, $d=0.63$ Adj. for IQ	
	GMDS Language	Adnams et al., (2001)	South Africa	68	7.12(0.5),	NR	FASD>CON, $p<0.001$ Adj. for IQ	
		Davies et al., (2017)	South Africa	121	NR	5-6	FASD=CON, $p=0.135$	
	Narrative Production	Thorne, (2017)	USA	138	9.78(NR)	7-12	Number of total words; FASD=CON Number of different words; FASD<CON, $p=0.0043$ mean length of utterance in morphemes; FASD<CON	
Memory *K*= 5, *n*= 399	Story Memory	Kalberg et al., (2013)	South Africa	113	6.54(0.63)	NR	FASD=CON, $p=0.318$ Adj. for age.	Children with FASD generally displayed poorer memory than non-exposed peers, including on assessments of immediate and delayed recall of visual information (Pei et al., 2011; Rasmussen, Pei, et al., 2009) and auditory recognition (Kully-Martens, Pei, Job, & Rasmussen, 2012). Children with FASD had greater prospective memory difficulties compared to non-exposed peers (Lewis et al., 2016). Children with FASD did not differ from peers on the story memory task in one study (Kalberg et al., 2013).
	Prospective Memory	Lewis et al., (2016)	South Africa	89	11(0.5)	NR	Focal cues; FASD<CON, $p<0.01$ Proximal cues; FASD<CON, $p<0.01$	
	Source Monitoring Task	Kully-Martens et al., (2012)	Canada	57	9.05(NR)	6-12	Overall recognition memory: FASD<CON, $p<0.05$, partial $\eta^2=0.085$ Source memory performance; FASD<CON, $p<0.05$, partial $\eta^2=0.113$	
	Memory Strategy task	Rasmussen, Pei, et al., (2009)	Canada	70	NR	5-10	Overall memory task performance: FASD<CON, $p<0.01$, $\eta^2=0.27$	
	Rey-Osterrieth Complex Figure Test	Pei et al., (2011)	Canada	70	8.29(NR)	6-12	Recall after 3 min; FASD<CON, $p=0.006$, $d=0.68$ Recall after 30 min; FASD<CON, $p=0.009$, $d=0.64$	
Motor Skills *K*= 12, *n*= 942	GMDS Motor Skills	Adnams et al., (2001)	South Africa	68	7.12(0.5),	NR	FASD=CON, p N.S Adj. for maternal education	Children with FASD showed mixed results on composite motor skills assessments (i.e., combined fine and gross assessments). Children with FASD had greater motor skills difficulties on the M-ABC and
		Davies et al., (2017)	South Africa	121	NR	5-6	FASD<CON, $p=0.035$	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
	Bruininks-Oseretsky Test of Motor Proficiency (BOT-2)	Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	Total scores; FASD=CON, p=0.36 Adj. for IQ	the COMPS (Kooistra et al., 2009), but not the BOT-2 Total scores. Mixed results were reported on the GMDS Motor Skills subscales, with one study reporting greater impairment (Davies et al., 2017) but no differences in an earlier study (Adnams et al., 2001). Mixed results were reported on gross motor skills assessments. Children with FASD had more impaired static and dynamic balance, but not ball skills on the M-ABC (Kooistra et al., 2009). No differences were reported between FASD and non-exposed peers on the BOT-2 gross motor composite scores (Lucas, Latimer, Doney, et al., 2016). Children with FASD were consistently noted to have difficulties with fine motor skills, as measured by the BOT-2, and the M-ABC (Doney, Lucas, Watkins, et al., 2017; Kooistra et al., 2009). On tests more specifically examining hand-writing skills, children with FASD had fewer legible words in each sentence compared to peers (Doney, Lucas, Jirikowic et al., 2017) On tests of visual-motor processing such as the VMI and the NEPSY visuospatial processing, children with FASD performed more poorly than peers in most studies (Coles et al., 1997; Kalberg et al., 2013; May et al., 2014). Although only one study provided the VMI subscale scores, contrasting results were noted between subscales, such that children with FASD scored more poorly on the motor coordination subscale compared to the visual perception subscale (Doney et al., 2016). Two studies reported sensorimotor measures (the QNST-II and the NEPSY Sensori-motor). Both studies reported children with FASD to have greater
		Doney, Lucas, Watkins, et al., (2017)	Australia	99	8.7(NR)	7-9	Fine motor composite; FASD<CON, p=0.046, η^2=0.06	
		Lucas, Latimer, Doney, et al., (2016)	Australia	99	8.7(0.6)	7-9	Gross motor composite; FASD=CON, p=0.32	
	Clinical Observation of Motor Skills (COMPS)	Kooistra et al., (2009)	Canada	116	8.8(1.2)	7-10	FASD<CON, p<0.001	
	Movement Assessment Battery for Children	Kooistra et al., (2009)	Canada	116	8.8(1.2)	7-10	Total Scores; FASD<CON, p<0.001 Manual Dexterity; FASD<CON, p<0.001 Static and Dynamic Balance; FASD<CON, p<0.001 Ball Skills; FASD=CON p N.S	
	Beery-Buktenica Developmental Test of Visual Motor Integration (VMI)	Coles et al., (1997)	USA	149	7.46(0.49)	7-8	Total Scores; FASD<CON, p<0.005	
		Kalberg et al., (2013)	South Africa	113	6.54(0.63)	NR	Total Scores; FASD<CON, p<0.005	
		May et al., (2014)	USA	216	6.96(0.54)	NR	Total Scores; FASD<CON, p<0.005	
		Doney et al., (2016)	Australia	99	8.7(NR)	7-9	Total scores; FASD=CON, p=0.091, η^2=0.05 Visual Perception; FASD=CON, p=0.229, η^2=0.03 Motor Coordination; FASD<CON, p=0.02, η^2=0.08	
	NEPSY Visuospatial processing	Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	FASD=CON, p=0.07 Adj. for IQ	
	NEPSY Sensori-Motor	Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	FASD<CON, p<0.001 Adj. for IQ	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
	Quick Neurological Screening Test-II (QNST-2)	Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	FASD>CON, $p<0.01$ Adj. for IQ	difficulties in this domain (Jirikowic, Olson, et al., 2008; Lucas, Latimer, Fitzpatrick, et al., 2016).
		Lucas, Latimer, Fitzpatrick, et al., (2016)	Australia	99	8.7(NR)	7-9	FASD>CON, $p<0.004$	
	Evaluation Tool of Children's Handwriting	Doney, Lucas, Jirikowic, et al., (2017)	Australia	99	8.7(NR)	7-9	Name letter legibility; FASD=CON, $p=0.729$ Sentence letter legibility; FASD=CON, $p=0.872$ Sentence word legibility; FASD<CON, $p=0.008$	
	Miller Function and Participation Scales: Draw a kid game	Doney, Lucas, Jirikowic, et al., (2017)	Australia	99	8.7(NR)	7-9	FASD=CON, $p=0.522$	
Social Skills, and Social Communication *K*= 9, *n*= 380	Social Skills Improvement System (SSIS) and Social Skills Rating Systems (SSRS)	Greenbaum et al., (2009)	Canada	97	9.23 (NR)	6-13	Parent/caregiver rated social skills; FASD<CON, $p<0.001$ Teacher rated social skills; FASD<CON, $p<0.001$ Parent/caregiver rated problem behaviour; FASD>CON, $p<0.001$ Teacher rated problem behaviour; FASD>CON, $p<0.001$ Adj. for abuse history, SES, IQ	Children with FASD had greater problem behaviours in social settings (SSIS problem behaviour) as rated by parents and teachers (Greenbaum et al., 2009; Jirikowic, Olson, et al., 2008; Stevens et al., 2015; Stevens et al., 2013). While parents consistently rated children with FASD as having lower social skills than their non-exposed peers (Greenbaum et al., 2009; Stevens et al., 2015; Stevens et al., 2013), mixed results were reported by teachers with one study reporting lower social skills (Greenbaum et al., 2009), and one reporting contrasting results (Jirikowic, Olson, et al., 2008). Children with FASD were rated by caregivers as having lower levels of empathy compared to typically developing peers (Stevens et al., 2015). On measures of social cognition such as theory of mind and emotion recognition, there were mixed
		Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	Teacher rated social skills; FASD=CON, $p=0.51$ Teacher rated problem behaviour; FASD>CON, $p<0.001$ Adj. for IQ	
		Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	Parent/caregiver rated social skills; FASD<CON, $p<0.001$, $d=0.59$ Parent/caregiver rated problem behaviour; FASD>CON, $p<0.001$, $d=0.60$ Adj. for age, gender	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
		Stevens, Nash, Koren, & Rovet, (2013)	Canada	42	10.3(1.5)	8-12	Parent/caregiver rated social skills; FASD<CON, p<0.001, d=0.50 Parent/caregiver rated problem behaviour; FASD>CON, p<0.001, d=0.73	results between children with FASD and typically developing peers. While children with FASD generally did not differ from peers on a false-belief task (Greenbaum et al., 2009; Lindinger et al., 2016; Rasmussen, Wyper, et al., 2009), mixed results were noted on increasingly complex assessments of social cognition such as strategic control of emotion (Greenbaum et al., 2009; Stevens et al., 2015). Mixed results were noted in tests of emotion recognition, with children with FASD performing similarly to non-exposed peers on tasks where they could see a full face or had limited choices of answers (Greenbaum et al., 2009; Lindinger et al., 2016), but showing performance deficits on tasks with less information available such as the Reading the Mind in the Eyes Test (Lindinger et al., 2016; Stevens, Clairman, Nash, & Rovet, 2017; Stevens et al., 2015). In studies using direct observations of children's behaviour, children with FASD were reported to spend more time in the classroom either disengaged from the tasks or engaging in irrelevant behaviours (Olswang, Svensson, & Astley, 2010). Further, children with FASD showed greater variability in the time spent engaged on the assigned task or engaging in irrelevant behaviour across days (Kjellmer & Olswang, 2013).
	SSIS Empathy	Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	SSIS Empathy Subscale; FASD<CON, p<0.001, η^2=0.30 Adj. for age, gender	
	Index of Empathy	Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	Index of Empathy; FASD<CON, p<0.01, η^2=0.15 Adj. for age, gender	
	NEPSY Social Perception subtests	Lindinger et al., (2016)	South Africa	63	10.6(0.4)	9-11	NEPSY Affect recognition; FASD=CON, p=0.370 NEPSY Contextual; FASD=CON, p=0.303	
		Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	NEPSY Contextual; FASD<CON, p<0.05, η^2= 0.08 NEPSY Verbal; FASD<CON, p<0.001, η^2= 0.24 Adj. for age, gender	
	Reading the Mind in the Eyes Test:	Lindinger et al., (2016)	South Africa	63	10.6(0.4)	9-11	FASD<CON, p<0.001	
		Stevens et al., (2017)	Canada	56	9.88(1.3)	8-12	FASD<CON, p<0.001, η^2= 0.19 Adj. for age, gender	
	Minnesota Test of Affective Processing	Greenbaum et al., (2009)	Canada	97	9.23 (NR)	6-13	Affect choice; FASD<CON, p<0.05 Affect naming, affect match, and prosody congruence; FASD=CON, p NR Adj. for abuse history, SES, IQ	
	Test of Social Cognition	Greenbaum et al., (2009)	Canada	97	9.23 (NR)	6-13	False Belief; FASD=CON, p NR Interpretative Theory of Mind; FASD=CON, p NR Strategic Control of Emotion; FASD<CON, p<0.05 Adj. for abuse history, SES, IQ	
		Stevens et al., (2015)	Canada	58	10.03(1.40)	8-12	False Belief; FASD<CON, p<0.001, η^2=0.35 Strategic Control of Emotion; FASD<CON p<0.01, η^2=0.15	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)
							Personalised Thoughts; FASD=CON, *p* NR Personalised Emotion; FASD<CON $p<0.05$, $\eta^2=0.11$ Adj. for age, gender
	Hoogenhout & Malcom-Smith Social Cognition test battery	Lindinger et al., (2016)	South Africa	63	10.6(0.4)	9-11	First order false belief; FASD=CON, $p=0.574$ Second order false belief; FASD=CON, $p=0.121$ Strange Stories; FASD=CON, $p=0.354$ Faux pas detection; FASD=CON, $p=0.506$ Faux pas comprehension; FASD=CON, $p=0.065$
	False-Belief tests (Wimmer and Perner puppet stories)	Rasmussen, Wyper, et al., (2009)	Canada	53	6.47(1.21)	4-8	FASD=CON, $p=0.068$
	Lie-telling behaviour	Rasmussen, Talwar, Loomes, & Andrew, (2008)	Canada	47	See note below	4-8	Children with FASD were more likely to tell a lie about peeking at a hidden toy during the task ($p<0.05$)
	Classroom Social Communication Coding System (SCCS)	Olswang et al., (2010)	USA	24	9.2(1.1)	7-12	Time engaged in classroom behaviour Prosocial/engaged; FASD<CON, $p<0.001$, $d=2.3$ Irrelevant; FASD>CON, $p<0.001$, $d=1.7$ Disengaged; FASD>CON, $p<0.001$, $d=3.2$ Assertive, Hostile/Coercive or Adult-seeking**;** FASD=CON, *p* NR
		Kjellmer & Olswang, (2013)	USA	24	9.2(1.1)	7-12	Variability (Kjellmer & Olswang, 2013) Changeability; FASD>CON, $p<0.001$

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
							Stability (prosocial/engaged); FASD>CON, $p<0.01$, partial $\eta^2=0.49$ Stability (irrelevant); FASD>CON, $p<0.01$, partial $\eta^2=0.07$ Stability (passive/disengaged); FASD=CON, $p=0.41$, partial $\eta^2=0.55$	
Sleep *K*= 2, *n*= 483	Children's Sleep Health Questionnaire (CSHQ)	Chen, Olson, Picciano, Starr, & Owens (2012)	USA	451	7.5(2.2)	4-12	CSHQ Total scores FASD>CON, $p<0.01$	Children with FASD had a greater level of sleep-related behavioural difficulties compared to typically developing peers in two studies (Chen et al., 2012; Wengel et al., 2011). Further, children with FASD were reported to get fewer hours sleep on average (Chen et al., 2012).
		Wengel, Hanlon-Dearman, & Fjeldsted, (2011)	Canada	31	4.92(NR)	3-6	CSHQ Total scores FASD>CON, $p<0.01$ Adj. for age	
	Total hours of sleep	Chen et al., (2012)	USA	451	7.5(2.2)	4-12	FASD<CON, $p<0.001$.	
Sensory Processing *K*= 3, *n*= 105	Sensory Profile Measure	Jirikowic, Olson, et al., (2008)	USA	51	6.5(0.88)	5-8	FASD>CON, $p<0.001$ Adj. for IQ	Children with FASD had a greater level of sensory processing difficulties compared to non-exposed peers in two studies (Jirikowic, Olson, et al., 2008; Wengel et al., 2011). A third study (Hansen & Jirikowic, 2013) reported a greater proportion of children with FASD to exceed the clinical cut-offs on two sensory processing measures, but did not conducted any inferential statistical analysis, limiting the conclusions that can be drawn.
		Wengel et al., (2011)	USA	31	4.92(NR)	3-6	FASD>CON, $p<0.001$ Adj. for age	
	Short Sensory Profile	Hansen & Jirikowic, (2013)	USA	33	8.75(2.16)	5-11	FASD 90.9% above clinical (81.8% definite dysfunction, 9.1% some problems), 0% non-exposed group above cut offs.	
	Sensory Processing Measure- Home Scale	Hansen & Jirikowic, (2013)	USA	33	8.75(2.16)	5-11	FASD 81.8% above clinical (18.2% definite dysfunction, 63.6% some problems), 0% non-exposed group above cut offs.	
FASD-Specific Scales	Personal Behaviour Checklist-36	Kalberg et al., (2013)	South Africa	113	6.54(0.63)	NR	Parent rated FASD>CON, $p<0.001$ Adj. for age	Seven studies reported behavioural measures intended to reflect behaviours characteristic of FASD. Children with FASD scored high than non-exposed peers in all seven studies (Breiner, Nulman, & Koren, 2013; Kalberg et al., 2013; Kodituwakku, Coriale, et
		Kodituwakku, Coriale, et al., (2006)	Italy	82	6.3(0.46)	6-7	Parent rated FASD>CON, $p<0.001$	

Table 2.4 *Key findings of studies comparing children diagnosed with FASD to non-exposed peers*

Domain *K* studies *n participants*	Outcome	Citation	Country	N	Age Mean (SD)	Age range	Key findings (Analysis not adjusted for co-variates unless otherwise stated)	
K= 7, *n*= 1,025		May et al., (2007)	South Africa	218	7.81(0.86)	NR	FASD>CON, $p<0.001$	al., 2006; May et al., 2007; May et al., 2015; Quattlebaum & O'Connor, 2013; Viljoen et al., 2005)
		May et al., (2015)	USA	216	7.13(0.48)	NR	FASD>CON, $p<0.001$	
		Quattlebaum & O'Connor, (2013)	USA	125	8.18(2.10)	6-12	Parent rated FASD>CON, $p<0.001$, $d= 1.99$. Teacher rated FASD>CON, $p<0.001$, $d=0.94$ Adj. for IQ	
		Viljoen et al., (2005)	South Africa	210	6.54(0.64)	NR	Parent rated FASD>CON, $p<0.001$	
	Neurobehavioural Screening Tool	Breiner et al., (2013)	Canada	60	NR(NR)	4-6	FASD>CON, $p<0.0001$	

Table 2.5 *Key findings of studies comparing children with PAE to non-exposed peers*

Domain *K* studies, *n* participants	Outcome	Citation	Country	N	Age Mean(SD)	Range	Key findings *Subscales (if applicable); Group differences, p values, effect size (if reported)*	Summary
Academic Achievement *K*= 2, *n*= 125	Arithmetic composite (WRAT-R and WISC-III scores converted to Z scores and combined)	Crocker, Riley, & Mattson, (2015)	USA	56	10.02(1.86)	7-12	PAE<CON, $p<0.05$.	Children with PAE at varying amounts had impairments arithmetic in two studies (Crocker et al., 2015; Godel et al., 2000; and literacy outcomes in one study (Godel et al., 2000)). The contrasting findings, of no effect amongst children with PAE reported by Godel and colleagues (2000), appear to be driven by poorer performance by students across the board on the math problems and time problem subscales rather than by better relative performance of the PAE group.
	Brigance Literacy subscales	Godel et al., (2000)[a]	Canada	69	NR(NR)	NR	Word recognition, oral reading, comprehension, spelling, mean grade equivalent; PAE<CON, $p<0.01$	
	Brigance Arithmetic subscales	Godel et al., (2000)	Canada	69	NR(NR)	NR	Number recognition, money problems, mean grade equivalent; PAE<CON, $p<0.01$ Math problems, time problems; PAE=CON, *p* NR	
Adaptive Behaviour *K*= 1, *n*= 65	Vineland Adaptive Behaviour Scale (VABS)	Crocker, Vaurio, Riley, & Mattson, (2009)	USA	65	10.32(2.37)	6-13	Socialization; PAE>CON, $p<0.001$ Living skills; PAE>CON, $p<0.001$ Communication; PAE>CON, $p<0.001$	Children heavily exposed to alcohol prenatally had lower adaptive behaviour scores across all subscales of the VABS in one study (Crocker et al., 2009)
Affect Regulation *K*= 1, *n*= 130	Mental Health Computerised Diagnostic Schedule for Children (4th Ed).	(Walthall et al., 2008)	USA	130	8.50(1.53)	6-12	PAE was a significant predictor of Separation anxiety ($\beta=0.24$, $p<0.05$); Generalised Anxiety Disorder ($\beta=0.17$, $p<0.05$);Externalising disorder: ADHD ($\beta=0.50$, $p<0.001$), Oppositional Defiant Disorder ($\beta=0.53$, $p<0.001$), Conduct disorder ($\beta=0.24$, $p<0.05$); Depression (not sig, after controlling co-variates); Symptoms of mania and hypo mania ($\beta=0.30$, $p<0.01$). PAE was not significantly related to Obsessive Compulsive Disorder, Post-Traumatic Stress Disorder, dysthymic disorder or schizophrenia.	PAE was associated with symptoms of anxiety, and conduct disorder but not depression (Walthall et al., 2008).
Attention *K*= 1, *n*= 69	ACTeRS Attention subscale	Godel et al., (2000)	Canada	69	NR(NR)	NR	PAE<CON, $p<0.001$	Children with PAE were rated by teachers has having greater attention difficulties on an indirect measure of

Table 2.5 *Key findings of studies comparing children with PAE to non-exposed peers*

Domain *K* studies, *n* participants	Outcome	Citation	Country	N	Age Mean(SD)	Range	Key findings *Subscales (if applicable); Group differences, p values, effect size (if reported)*	Summary
								attention, compared to non-exposed peers (Godel et al., 2000).
Executive function, inc. hyperactivity and impulsivity *K*= 4, *n*= 237	Fluency	Vaurio, Riley, & Mattson, (2008)	USA	60	10.68(1.65)	7-13	Letter fluency; PAE<CON, $p<0.05$ Category fluency; PAE=CON, $p=0.059$	Children with PAE appeared to have a greater level of executive functioning impairment across fluency (Vaurio et al., 2008), set-shifting (Vaurio et al., 2008), problem solving (McGee, Bjorkquist, Price, Mattson, & Riley, 2009) and working memory (Crocker et al., 2015). Further, children with PAE were rated by teachers as displaying a greater level of hyperactive behaviours compared to non-exposed peers (Godel et al., 2000).
	Trail-making Test	Vaurio et al., (2008)	USA	60	10.68(1.65)	7-13	Form A (Consecutive); PAE=CON, $p>0.10$ Form B (Switching); PAE<CON, $p=0.023$	
	Wisconsin Card Sorting Test (WCST)	Vaurio et al., (2008)	USA	60	10.68(1.65)	7-13	PAE<CON, $p<0.01$ on all WCST indices	
	Test of Problem Solving	McGee, Bjorkquist, Price, et al., (2009)	USA	52	9.42(1.58)	7-11	PAE<CON, $p<0.001$, $d=0.99$	
	Digit Span	Crocker et al., (2015)	USA	56	10.02(1.86)	7-12	PAE<CON, $p<0.05$	
	Spatial Span	Crocker et al., (2015)	USA	56	10.02(1.86)	7-12	PAE<CON, $p<0.05$.	
	ACTeRS Hyperactivity subscale	Godel et al., (2000)	Canada	69	NR(NR)	NR	PAE>CON, $p<0.001$	
Language and Communication *K*= 1, *n*= 51	Clinical Evaluation of Language Fundamentals-Preschool (CELF-P	McGee, Bjorkquist, Riley, & Mattson, (2009)	USA	51	4.35(0.85)	3-5	Total language composite scores: PAE<CON, $p<0.001$, $d=0.98$ Receptive language: PAE<CON, $p<0.001$, $d=0.96$ Expressive language: PAE<CON, $p<0.001$, $d=0.82$	Children with PAE appeared to have greater difficulties with both expressive and receptive language (McGee, Bjorkquist, Riley et al., 2009).
Memory *K*= 1, *n*= 56	CANTAB Spatial Recognition	Crocker et al., (2015)	USA	56	10.02(1.86)	7-12	Spatial Recognition Memory; PAE<CON, $p<0.05$ Pattern Recognition Memory; PAE=CON, p N.S	Children with PAE had greater levels of impairment on spatial recognition memory (Crocker et al., 2015).
Social skills *K*= 3, *n*= 142	Social Skills Rating System (SSRS)	Rasmussen, Becker, McLennan, Urichuk, &	Canada	60	5.1(1.6)	3-8	Parent/caregiver rated social skills; PAE<CON, $p<0.05$, partial $\eta^2=0.08$ Teacher rated social skills; PAE=CON, $p=0.57$. Parent/caregiver rated problem behaviour; PAE=CON, $p=0.72$	Mixed results noted amongst social skills between children with PAE and CON. Children with PAE were rated by their parents/caregivers as having poorer social skills than typically

Table 2.5 *Key findings of studies comparing children with PAE to non-exposed peers*

Domain *K* studies, *n* participants	Outcome	Citation	Country	N	Age Mean(SD)	Range	Key findings *Subscales (if applicable); Group differences, p values, effect size (if reported)*	Summary
		Andrew, (2011)					Teacher rated problem behaviour; PAE=CON, p=0.60.	developing peers recruited from a behavioural support program (Rasmussen et al., 2011). No differences were noted on teacher-rated social skills, or parent/caregiver- or teacher- rated behavioural concerns. Children with PAE had greater difficulties on a number of the play skills evaluated by Pearton and colleauges (2014). Lastly, children with PAE were reported to display difficulties in identifying effective responses to hostile behaviour in social situations, had difficulties attributing intent, and had greater difficulty in identifying whether their response in a social situation had the desired outcome (McGee, Bjorkquist et al., 2009).
	Test of Playfulness	Pearton, Ramugondo, Cloete, & Cordier (2014)	South Africa	30	7.05(0.5)	NR	PAE<CON on the following Test of Playfulness subscales: Engages in Social Play, intensity (p<0.01); Engages in Social Play, skill (p<0.04); Initiates play with others (p<0.01); Clowns or jokes, extent (p<0.01); Clowns or jokes, skill (p<0.03). PAE=CON (p>0.05) on the following subscales on the extent with which engages in social play and the skills with which the negotiates with others to have needs met, supports the play of others, enters a group already engaged in activity, shares, gives readily understandable cues, and responds to other's cues.	
	Social Information Processing	McGee, Bjorkquist, Price, et al., (2009)	USA	52	9.42(1.58)	7-11	Children with PAE displayed difficulties with identifying effective responses in social situations, attribution of intent, and in evaluating the response to their actions.	
FASD Specific-scales *K*= 1, *n*= 69.	FAS/FAE Score	Godel et al., (2000)	Canada	69	NR(NR)	NR	PAE>CON, p<0.001	Children prenatally exposed to alcohol had a higher score than than peers on a behavioural checklist, created to measure the behavioural profile of children with FASD (Godel et al., 2000).

Table 2.6 *Key findings of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol*

Domain	Cohort	Study citations	Outcomes (Subscale in parentheses where appropriate)	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
Academic achievement	ALSPAC	Alati et al., (2013)	Key Stage 2 (KS2) Academic Achievement	11 (*n*=7,062)	Regular consumption categories: never, <1 per week, 1-6 a week, 1+ a day, Binge drinking frequency (days over pregnancy): None, 1-4 days, 5-10 days, 10+ days).	Increase in frequency of binge drinking was associated with lower mean KS2 Scores, indicating poorer academic achievement. Change in mean scores, after adjusting for covariates= -0.68(95% CI=-1.03- -0.33, *p*<0.0001).	Exposure to binge drinking was associated with poorer academic achievement at age 11 in the UK ALSPAC studies (Alati et al., 2013; Sayal et al., 2014). Heavy alcohol exposure was also associated with an increased risk of not achieving national benchmarks in reading or writing, but not for maths in the Australian RASCALS cohort (O'Leary et al., 2013). High levels of alcohol exposure (1.5 oz AA/day) was associated with lower maths skills in one study (Larroque & Kaminski, 1998) Low levels of exposure were not associated with lower education outcomes in the ALSPAC cohort (≥1 drink per week; Sayal et al., 2013), or the UK Millenium cohort (1-2 drinks per week; Kelly et al., 2013).
		Sayal et al., (2013)	KS2	11 (*n*=10,558)	None, ≤1 drink per week, ≥1 drink per week	No effect of light alcohol (≥1 drink per week) mean academic achievement scores (change in KS2 scores= 0.45 (95% CI=0.11-1.01)), after adjusting for covariates.	
		Sayal et al., (2014)	KS2	11 (*n*=6,939)	At least one occasion of binge drinking (>4 drinks in a day)	Binge drinking was associated with lower academic achievement (Adjusted mean difference= -0.81 (95% CI=-0.16- -1.46).	
	RASCALS	O'Leary et al., (2013)	Western Australian Literacy and numerary assessment	10 (*n*=4,056)	None, Low (1-2 drinks/session, <7/wk), moderate (3-4 drinks/session, =7/wk), occasional binge (5+ drinks/session, less than weekly, heavy (>7/wk, including binge drinking weekly or more frequently)	Children of mothers who drank heavily in the first trimester had increased risk of not achieving the benchmark for reading (aOR=2.27, 95% CI=1.11-4.67). Further, occasional binge drinking in late pregnancy increased the odds of not achieving the benchmark for writing (aOR=2.35, 95% CI=1.04-5.43).	
	Millenium	Kelly et al., (2013)	British Assessment Scale (BAS) Reading assessment NFER Progress in Maths test	7 (*n*=9,597)	Groups other than those reported no alcohol consumption in pregnancy and those who drank "1-2 units per week or per occasion during pregnancy"	No increased risk of negative outcomes following light PAE on the behavioural or cognitive outcomes. Small protective effect of light alcohol consumption on reading and spatial skills amongst boy, but not girls.	
	Roubaix birth cohort	Larroque & Kaminski, (1998)	McCarthy Scales Quantitative subscales	4.5 (*n=160*)	Consumptions of 1.5oz absolute alcohol or more per day during pregnancy	PAE<CON, *p*=0.001	
Affect regulation	DNBC	Niclasen, Andersen, Strandberg-Larsen, et al., (2014)	SDQ Emotional Problems, SDQ Peer problems	7 (*n*=37,315)	No reports of binge drinking, and reports of binge drinking in 1st (Early group) or 3rd (Late group) trimester. [Note: all other possible groups excluded]	No difference in SDQ emotional and peer problems scores were reported between groups exposed in either early or late pregnancy.	Exposure to binge drinking was associated with an increased risk of internalising behaviour, as measured by the SDQ in the DNBC cohort (Niclasen, Andersen, Teasdale et al., 2014), and the SDQ emotional problems subscale in the Olso Pregnancy Cohort (Alvik et al., 2013). Mixed results were noted across two studies which focused on binge exposure in the early stages of pregnancy; whereas Alvik and colleagues (2013) reported an increased risk of high emotional problem scores on the SDQ following binge exposure in the first 6 weeks of pregnancy, Niclasen, Andersen, Strandberg-
		Niclasen, Andersen, Teasdale, et al., (2014)	SDQ (Emotional, peer problems, internalising scale)	7 (*n*=37,152)	Binge drinking frequency 0, 1, 2-3, 4+ binge episode in pregnancy, and cumulated total number of drinks 0, 0-5, 5-15, 15-45, 45-90, 90+ (90+= >2.4 per week)	Binge drinking was associated with increased risk of SDQ internalising (relative change in mean = 1.03-1.07) for boys. Apparent protective factor of high average exposure (90+ cumulated drinks) on emotional problems for boys (OR:	

Table 2.6 *Key findings of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol*

Domain	Cohort	Study citations	Outcomes (Subscale in parentheses where appropriate)	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
						0.71 (CI: 0.59-0.85)) and girls (OR: 0.82 (CI: 0.69-0.98)).	Larsen et al., (2014) reported both no increased risk of high emotional problems scores, or higher mean scores among children exposed to binge drinking in the first trimester.
	RASCALS	O'Leary et al., (2010))	CBCL	8 (*n*=1,846)	None, low (<1 drink/day), moderate (1 drink/day), heavy (>1 drink/day).	Low levels of PAE were not associated with increased risk of child behaviour problems. Increase risk of anxiety/depression (aOR=2.82, 95% CI=1.07-7.43), somatic complaints (aOR=2.74, 95% CI=1.47-5.12), following heavy alcohol exposure.	
	Oslo Pregnancy Cohort	(Alvik, Aalen, & Lindemann, 2013)	SDQ (Emotional) Likelihood of scoring in borderline, or abnormal range	5 (*n*=1,003)	Binge drinking in very early (weeks 0-6) of pregnancy-never, less than once a week, more than once a week.	Children exposed to early binge drinking had increased odds of obtaining a high symptom score on the SDQ emotional problems (OR=2.8, $p<0.05$)	
Attention	DNBC/ LSDP	(Kesmodel et al., 2012)	Test of Everyday Attention (TEACH-5)	5 (*n*=1,628)	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+) Binge drinking= dichotomised to any or none	No statistically significant effect of weekly alcohol consumption, or binge drinking TEACH-5 adjusted mean difference= -0.01 (95% CI=-0.14-0.11).	Children exposed to 9 or more drinks per week throughout pregnancy were found to have a greater risk of achieving a below-average score, defined as more than 1 SD below the group mean on the TEACH at 5 years old (Underbjerg et al., 2012). In the same cohort, there was no difference on mean scores across the different average exposure groups, or between those exposed to any incidents of binge drinking (Kesmodel et al., 2012).
		(Underbjerg et al., 2012)	TEACH-5 (overall score, and risk of below average score)	5 (n=1,617)	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+) Binge drinking variables included yes/no at any point, total number 0, 1, 2, 3+	Heavy alcohol exposure (9+) was associated with an increased risk of a below average score (>1SD below mean) (aOR=3.20, 95% CI=1.08-9.53). No associations were found between lower exposures categories and attention outcomes.	
	Gesta-Alcohol Study, Brazil	Furtado & Roriz, (2016)	d2 Test of Attention	11-12 (*n*=56)	Any positive answer to alcohol consumption questions (e.g., quantity or frequency, or a score above one on the T-ACE, or a positive ICD-10 diagnosis for harmful alcohol use or dependence	Rough result (speed performance); PAE<CON, $p=0.01$ Total errors (impulsivity); PAE<CON, $p=0.02$ Net results (focused attention); PAE=CON, $p=0.14$	Compared to non-exposed peers, children aged 11 to 12 years old with any reports of PAE had lower speed perform and greater impulsivity that non-exposed peers. No differences were reported between children with PAE and non-exposed peers on focused attention (Furtabo & Roriz, 2016).
	Finland	Korkman et al., (1998)	Attention composite (battery of inhibition tasks and sustained attention task)	5-9 (*n=72*)	More than 10 drinks per week, split by trimesters; 1) ceased alcohol exposure after first trimester, 2) alcohol exposure in first and second trimester, 3) alcohol exposure throughout pregnancy, and 4) non-exposed comparison	PAE<CON, $p=0.01$	Children exposed to alcohol throughout pregnancy had lower scores on an attention composite, compared to non-exposed peers (Korkman et al., 1998)
Executive functioning	ALSPAC	(Sayal et al., 2013)	SDQ Conduct, (Hyperactivity / inattention, total problems)	11 (*n*=6,587)	None, ≤1 drink per week, ≥1 drink per week	No effect of light alcohol (≥1 drink per week) consumption on teacher-rated SDQ scores (change in SDQ total problem score= -0.05 (95% CI=-017-0.07)	Binge drinking associated with greater levels of hyperactivity and impulsivity (Sayal et 2009; Sayal et al., 2014; Niclasen Andersen, Strandberg-Larsen et al., 2014; Niclasen, Andersen, Teasdale et al., 2014).
		(Sayal et al., 2014)	SDQ	11 (*n*=4,610)	At least one occasion of binge drinking (>4 drinks in a day)	Binge drinking was associated with higher levels of hyperactivity and inattention (adjusted mean difference= 0.28 (95% CI 0.04, 0.51)	

Table 2.6 *Key findings of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol*

Domain	Cohort	Study citations	Outcomes (Subscale in parentheses where appropriate)	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
			(Conduct, Hyperactivity / inattention, total problems)				Binge drinking or average alcohol consumption was not associated with elevated scored on the BRIEF (Kesmodel, 2012). Any alcohol consumption was associated with lower scores on category fluency, compared to non-exposed peers (Furtado et al., 2016). No differences were noted on letter fluency or set shifting as measured by the WCST.
		(Sayal et al., 2009)	SDQ (Conduct, Hyperactivity / inattention, total problems)	3.4 (*n*=6355) 6.8 (*n*=5599)	At least one occasion of binge drinking (>4 drinks in a day)	Binge drinking was associated with higher risk for hyperactivity/ impulsivity (adjusted mean difference= 0.25 (95% CI=0.11=0.40), and greater total problems (Adjusted mean difference= 0.46 (95% CI=0.17- 0.74).	
		(Sayal et al., 2007)	SDQ (Conduct, Hyperactivity / inattention, total problems)	3.4 (*n*=9,086) 6.8 (*n*=8,046)	None, ≤1 drink per week, ≥1 drink per week	High SDQ scores were not adequately defined in this study. Drinking ≤1 glass per week in the first trimester was associated with high total teacher SDQ scores (aOR: 1.36; 95% CI: 1.04–1.78; *p*=.025).	
	DNBC	(Niclasen, Andersen, Strandberg-Larsen, et al., 2014)	SDQ (Hyperactivity and conduct)	7 (*n*=37,315)	No reports of binge drinking, and reports of binge drinking in 1st (Early group) or 3rd (Late group) trimester. [Note: all other possible groups excluded]	Binge drinking in early or late pregnancy associated with slightly higher mean SDQ hyperactivity (1.02 (95% CI=1.00–1.05)); and conduct (1.21 (95% CI=1.04–1.42) subscale scores. No change for internalizing scores (emotional and peer problems) were reported.	
		(Niclasen, Andersen, Teasdale, et al., 2014)	SDQ (Hyperactivity and conduct subscales)	7 (*n*=37,152)	Binge drinking frequency 0, 1, 2-3, 4+ binge episode in pregnancy, and cumulated total number of drinks 0, 0-5, 5-15, 15-45, 45-90, 90+ (90+= >2.4 per week)	Binge drinking was associated with increased risk of SDQ internalising (relative change in mean = 1.03-1.07) and externalising (relative change in mean = 1.01-10.7) for boys. Binge drinking overall associated with conduct scores above cut-offs (OR: 1.12-1.23) for boys. Apparent protective factor of high average exposure (90+ cumulated drinks) on inattention for boys (OR: 0.79 (CI: 0.65-0.96)), and emotional problems for boys (OR: 0.71 (CI: 0.59-0.85)) and girls (OR: 0.82 (CI: 0.69-0.98)).	
		(Skogerbo et al., 2013)	SDQ (Total problems)	5 (*n*=1,617)	0, 1-5, 5+ drinks per week Number of binge episodes 0, 1,2, 3+	No association was noted between average drinks/week, (aOR=1.1, 95% CI=0.5–2.3) and aOR 1.1., 95% CI=0.6–2.1 for the total difficulties scores) or binge drinking episodes, and SDQ scores in the borderline or abnormal range, as rated by parents or teachers.	
		(Kesmodel et al., 2012)	Behavioural Rating Inventory of Executive Functioning (BRIEF)	5 (*n*=1,628)	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+) Binge drinking= dichotomised to any or none	No statistically significant effect of weekly alcohol consumption, or binge drinking on the BRIEF (adjusted mean difference= 0.17 (95% CI=1.01-1.35)	
	RASCALS	(O'Leary et al., 2010))	CBCL	8 (*n*=1,846)	None, low (<1 drink/day), moderate (1 drink/day), heavy (>1 drink/day).	Low levels of PAE were not associated with increased risk of child behaviour problems. Increase risk of anxiety/depression (aOR=2.82, 95% CI=1.07-7.43), somatic complaints	

Table 2.6 *Key findings of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol*

Domain	Cohort	Study citations	Outcomes (Subscale in parentheses where appropriate)	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
						(aOR=2.74, 95% CI=1.47-5.12), following heavy alcohol exposure.	
	Oslo Pregnancy Cohort	(Alvik, Aalen, & Lindemann, 2013)	SDQ (total problems, emotional and hyperactivity / impulsivity)	5 (*n*=1,003)	Binge drinking in very early (weeks 0-6) of pregnancy-never, less than once a week, more than once a week.	Children exposed to early binge drinking had an increased odds of obtaining a high symptom score on the SDQ total (OR=3.9, $p<0.01$), emotional problems (OR=2.8, $p<0.05$), conduct problems (OR=3.8, $p<0.01$), and hyperactivity /inattention (OR=3.8, $p<0.01$).	
	Millenium	(Kelly et al., 2013)	SDQ (Total problem scores)	7 (*n*=9,597)	Groups other than those reported no alcohol consumption in pregnancy and those who drank "1-2 units per week or per occasion during pregnancy" were excluded.	No increased risk of negative outcomes following light PAE on the behavioural or cognitive outcomes.	
	Gesta-Alcohol Study, Brazil	Furtado & Roriz, (2016)	Letter and Category fluency, Wisconsin Card Sorting Test	11-12 (*n*=56)	Any positive answer to alcohol consumption questions (e.g., quantity or frequency, or a score above one on the T-ACE, or a positive ICD-10 diagnosis for harmful alcohol use or dependence	Letter fluency; PAE=CON, $p=0.79$ Category fluency; PAE<CON, $p<0.05$ PAE=CON, $p>0.05$ on all WCST indices	
Language	Finland	Korkman et al., (1998)	Receptive language composite (auditory analysis, repeating non-words and the token test). Naming composite (naming colours, body parts, speeded naming and name learning).	5-9 (*n=72*)	More than 10 drinks per week, split by trimesters; 1) ceased alcohol exposure after first trimester, 2) alcohol exposure in first and second trimester, 3) alcohol exposure throughout pregnancy, and 4) non-exposed comparison	Receptive language PAE<CON, $p=0.01$ Naming measures PAE<CON, $p=0.01$	Children exposed to alcohol throughout pregnancy had lower scores on the receptive language composite and the naming composite compared to unexposed peers (Korkman et al., 1998).
Memory	Finland	Korkman et al., (1998)[1]	Verbal Memory composite (word span and logical learning) Visual Memory composite (memory for faces)	5-9 (*n=72*)	More than 10 drinks per week, split by trimesters; 1) ceased alcohol exposure after first trimester, 2) alcohol exposure in first and second trimester, 3) alcohol exposure throughout pregnancy, and 4) non-exposed comparison	Verbal memory PAE=CON, *p* N.S Visual memory PAE=CON, *p* N.S	Further, among children aged 11 to 12, prenatal alcohol exposure was associated with lower recall scores on the RCFT after 3 and 30 minutes (Furtado & Roriz, 2016). Contrasting results were found in other studies. Children exposed to alcohol throughout pregnancy did not differ from non-exposed peers on a verbal memory composite, or a visual memory composite. (Korkman et al., 1998). Further, children, aged 4 years old, exposed to 1.5 fl oz per day during pregnancy did not differ from peers on the McCarthy Memory Scales (Larroque & Kaminski, 1998).
	Gesta-Alcohol Study, Brazil	Furtado & Roriz, (2016)	Rey-Osterrieth Complex Figure Test (RCFT) Rey Auditory Verbal Learning Test	11-12 (*n*=56)	Any positive answer to alcohol consumption questions (e.g., quantity or frequency, or a score above one on the T-ACE, or a positive ICD-10 diagnosis for	Recall after 3 min: PAE<CON, $p=0.01$ Recall after 30 min: PAE<CON, $p=0.002$ Immediate recall, Recall after 30 min, Recognition, Proactive interference, Retroactive interference; PAE=CON, $p>0.05$	

Table 2.6 *Key findings of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol*

Domain	Cohort	Study citations	Outcomes (Subscale in parentheses where appropriate)	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
					harmful alcohol use or dependence		
	Roubaix birth cohort	Larroque & Kaminski, (1998)	McCarthy Scales Memory	4.5 (*n=160*)	Consumptions of 1.5oz absolute alcohol or more per day during pregnancy	PAE=CON, *p*=0.95	
Motor Skills	DNBC	(Bay et al., 2012)	Movement Assessment Battery for Children (M-ABC)	5 (*n*=685)	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+)	Alcohol consumption was not associated with motor skills as measured by the M-ABC at age 5, across average alcohol consumption categories 1-4 drinks/week (adjusted mean difference= 0.13 (95% CI=-1.39-1.66)), 5 to 8 drinks/week (-1.42 (95% CI=-3.83-0.98)) and 9+ drinks/week (0.64 (95% CI=-5.64-6.92)).	No difference in mean M-ABC scores was reported across groups of children exposed to light, moderate or heavy alcohol consumption in the ALPAC cohort (Bay et al., 2012). One study found alcohol use in pregnancy to be associated with greater scores on balance tasks at age ten (Humphriss et al., 2013). The authors, despite adjusting for a number of variables conclude that the findings may be accounted for by unmeasured co-variates.
	ALSPAC	(Humphriss et al., 2013)	Balance Tasks	10 (n=6,915)	Regular consumption per week: None, 1-2 (low), 3-7 moderate, 7+ high	Higher alcohol use in pregnancy was associated with better balance (aOR= 1.23, 95% CI=1.01-1.49)	
		Korkman et al., (1998)[1]	Visual Motor production composite (Visual Motor Integration and NEPSY Visuo-Motor Precision)	5-9 72	More than 10 drinks per week, split by trimesters; 1) ceased alcohol exposure after first trimester, 2) alcohol exposure in first and second trimester, 3) alcohol exposure throughout pregnancy, and 4) non-exposed comparison	PAE<CON, *p*=0.026	
		Korkman et al., (1998)	Manual Praxis (sensorimotor functions from the NEPSY; Fingertip Tapping, Imitating Hand Positions, Manual Motor Sequences)	5-9 72	More than 10 drinks per week, split by trimesters; 1) ceased alcohol exposure after first trimester, 2) alcohol exposure in first and second trimester, 3) alcohol exposure throughout pregnancy, and 4) non-exposed comparison	PAE=CON, *p.* NR	
	Roubaix birth cohort	Larroque & Kaminski, (1998)	Minor Neurological Anomalies	4.5 160	Consumptions of 1.5oz absolute alcohol or more per day during pregnancy	PAE>CON, *p*=0.002	
	Roubaix birth cohort	Larroque & Kaminski, (1998)	McCarthy Scales Motor Skills	4.5 160	Consumptions of 1.5oz absolute alcohol or more per day during pregnancy	PAE=CON, *p*=0.65	
Sleep	Finnish Birth Cohort	(Pesonen et al., 2009)	Sleep Disturbance Scale for Children	8 (*n*=289)	Number of drinks per week, dichotomised to drinking more than 1 standard drink per week	Children exposed prenatally to alcohol had an increased risk for short sleep duration (aOR=2.9, 95% CI=1.1-7.6) and low sleep efficiency (aOR=3.6, 95% CI=1.3-10.0).	Children exposed to more than one drink per week had an increased risk of short sleep duration, and low sleep efficiency (Pesonen et al., 2009).

Table 2.7 *Key findings of longitudinal studies examining the relationship between cognitive and behavioural outcomes and alcohol exposure as a continuous variable*

Domain *K* studies	Cohort	Study citations	Outcomes	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
Academic achievement	Detroit Sept 1986-Aug 1989 cohort	(Burden, Jacobson, Sokol, et al., 2005)	WISC-III Arithmetic	7.5 (*n*=337)	Average alcohol consumption per day during pregnancy	Arithmetic (β=-.15, *p*<0.01), was negatively related to PAE, after controlling for co-variates.	Average alcohol exposure per day during pregnancy was negatively associated with arithmetic in two studies (Burden, Jacobson, Sokol et al., 2005; Goldschmidt et al., 1996), and spelling and reading in one study (Goldschmidt et al., 1996).
	Maternal Health Practices	(Goldschmidt et al., 1996)	Wide Range Achievement Test-Revised (WRAT-R)	6.5 (*n*=512)	Average number of drinks per day during pregnancy, split by trimester	Each additional drink per day during pregnancy was associated with poorer performance on the arithmetic (β=-3.0, *p*<0.01), spelling (β=-2.5, *p*=0.03), and reading (β=-2.1, p=0.03) scales of the WRAT-R.	
Affect regulation	Clinic-referred	(O'Connor & Paley, 2006)	Pictorial Depression Scale	4.5 (*n*=42)	Retrospective report of prenatal alcohol consumption and defined as the maximum number of drinks per drinking occasion	Alcohol exposure was a significant predictor of negative affect, after controlling for confounding variables (β=0.36, *p*<0.05). Using bi-variate correlation, (and not controlling for other variables), PAE was associated with higher depressive symptoms (*r*=0.35, *p*<0.05).	Alcohol exposure was associated with symptoms of depression (Walthall et al., 2006, O'Connor & Paley, 2006) and anxiety (Walthall et al., 2008).
	Clinic-referred	(Walthall et al., 2008)	Mental Health Computerised Diagnostic Schedule for Children (4th Ed).	6-12 (*n*=130)	Caregivers completed the "Health interview for Women" when their children were between 6 and 12 years of age, and PAE was defined as a dichotomised exposed or non-exposed without additional information provided. The authors note that medical or legal records and collateral reported were sought to assess PAE amongst children in foster or adoptive care.	PAE was a significant predictor of Separation anxiety (β=0.24, p<0.05); Generalised Anxiety Disorder (β=0.17, p<0.05);Externalising disorder: ADHD (β=0.50, p<0.001), Oppositional Defiant Disorder (β=0.53, p<0.001), Conduct disorder (β=0.24, p<0.05); Depression (not sig, after controlling co-variates); Symptoms of mania and hypo mania (β=0.30, p<0.01). PAE was not significantly related to Obsessive Compulsive Disorder, Post-Traumatic Stress Disorder, dysthymic disorder or schizophrenia.	
Attention	Detroit Sept 1986-Aug 1989 cohort	(Burden, Jacobson, Sokol, et al., 2005)	Sustained attention composite made up of CPT,	7.5 (*n*=337)	Average alcohol consumption per day during pregnancy	The focus-sustain composite showed a weak negative relationship with average alcohol consumption per day.	Mixed results were observed on between measures of attention and alcohol exposure measured as a continuous variable. Alcohol exposure was associated with poorer attention as measured by direct assessment in several studies (Burden, Jacobson, Sokol et al., 2005; Chiodo et al., 2010; Chiodo 2009). Several studies identified no relationship between alcohol exposure and direct assessments of attention (Richardson et al., 2002), or parent-reported attention problems (D'Onofrio et al., 2007).
	Detroit Sept 1989- Aug 1991 cohort	(Chiodo et al., 2010)	CPT TRF Attention Scores	7 (*n*=462)	Average alcohol consumption per day during pregnancy, and quantity of alcohol per drinking day (binge)	Negative associations between prenatal binge drinking and a measure of impulsive responding on the CPT (β=-.63, *p*<0.01,), after adj. for covariates, but not average alcohol per day. No other associations were reported.	
	Maternal Health Practices	(Richardson et al., 2002)	CPT	10 (*n*=593)	Average number of drinks per day during pregnancy, split by trimester	No associated was reported between average alcohol exposure per day during pregnancy or binge drinking on the CPT.	

Table 2.7 *Key findings of longitudinal studies examining the relationship between cognitive and behavioural outcomes and alcohol exposure as a continuous variable*

Domain *K* studies	Cohort	Study citations	Outcomes	Age (*N* participants)	Alcohol exposure definitions	Findings	Summary
	Norwegian Mother and Child Birth Cohort Study	(Eilertsen et al., 2017)	Conner's Parent Rating Scale (CPRS-R) CBCL: ADHD Sub-scale	5 (n=114,247)	AUDIT-C scores based on first trimester drinking behaviour used as alcohol exposure measure. (Summed to a score for 0-12)	No relationship between PAE and ADHD diagnosis (Hazard Ratio=0.97, 95% CI=0.93-1.01). After adjusting for co-variates, PAE was associated with ADHD symptoms as measured by the CBCL (β=-0.040, 95% CI=0.024-0.055) and the CPRS-R (β=0.054, 95% CI=0.024-0.055).	
	Wayne State University (Detroit, MI) between February 1999 and January 2001	(Chiodo et al., 2009)	CPT Divided Attention	4.5 (*n*=75)	Defined by score on "At-risk alcohol exposure metric" Combination of average alcohol/day at conception, first perinatal visit, and across pregnancy, average drinks per drinking day, and standardised alcohol screeners such as the T-ACE.	At Risk Alcohol Exposure (ARAE) was negatively associated Divided attention (β=-0.26, p<0.025).	
	National Longitudinal Survey of Youth	(D'Onofrio et al., 2007)	CBCL attention/ impulsivity problems	4-11 (*n*=8,621)	Number of drinks per week, recalculated from 1/mo, 1/mo, 3-4 d/mo, 1-2 d/wk, 3-4 d/wk Nearly every day, Everyday	Drinks/week was not associated with attention problems after controlling for co-variates (*b*=0.02, *p*=0.24).	
Executive function	Maternal Health Practices	(Richardson et al., 2002)	WCST, Trail-Making, Stroop Task	10 (*n*=593)	Average number of drinks per day during pregnancy, split by trimester	No significant associations were noted between average alcohol exposure per day and executive function outcomes (WCST, Trail-Making test, Stroop test).	Mixed results on measures of executive functioning were reported. While several studies identified negative relationships between average alcohol consumption per day and executive functioning outcomes (Burden, Jacobson, Sokol, et al., 2005; Carter et al., 2016), conflicting results were identified by Richardson et al., (2002), who reported no relationship between average drinks per day and executive functioning outcomes. Prenatal alcohol exposure was associated with greater externalising behaviour problems as rated by teachers (Bailey et al., 2004) and parents (D'onofrio et al., 2007).
	Cape Town	(Carter et al., 2016)	DKEFS Fluency	5, 9 (*n*=157)	Averaging >1.0 oz absolute alcohol (Oz AA) per day (~2 standard drinks) or at least 2 binge episodes in the first trimester.	PAE (Oz AA/day) was associated with DKEFS Fluency category switching (*r*=-0.24, *p*<0.01).	
	Detroit Sept 1986-Aug 1989 cohort	(Burden, Jacobson, Sokol, et al., 2005)	Wisconsin Card Sorting Test (WCST) Tower of London WISC-III, digit span, category fluency	7.5 (*n*=337)	Average alcohol consumption per day during pregnancy	Working memory tests (digit cancelation β=.11, *p*<0.05), digit span β=-0.17, *p*<0.01, tower of London β=-.13, *p*<0.05, were negatively related to PAE, after controlling for co-variates.	
	Detroit Sept 1989, August 1991	(Bailey et al., 2004)	TRF	7 (*n*= 556)	Average alcohol consumption per day, and binge drinking (defined as present if > 5 drinks per occasions at least once every two weeks).	Exposure to binge drinking was associated with greater externalising behaviours, as rated by teachers.	

Table 2.7 *Key findings of longitudinal studies examining the relationship between cognitive and behavioural outcomes and alcohol exposure as a continuous variable*

Domain *K* studies	Cohort	Study citations	Outcomes	Age *(N* participants)	Alcohol exposure definitions	Findings	Summary
	National Longitudinal Survey of Youth	(D'Onofrio et al., 2007)	CBCL- conduct problems	4-11 (*n*=8,621)	Number of drinks per week, recalculated from 1/mo, 1/mo, 3-4 d/mo, 1-2 d/wk, 3-4 d/wk Nearly every day, Everyday [See D'Onofrio et al., 2007 for full conversion details].	Drinks/week was significantly, positively associated with conduct problems (*b*=0.05, *p*=0.04),	
Memory	Maternal Health Practices	(Richardson et al., 2002)	WCST WRAML Trail-Making Stroop Task Grooved Pegboard CPT	10 (*n*=593)	Average number of drinks per day during pregnancy, split by trimester	First trimester alcohol consumption predicted performance on WRAML Design memory, verbal learning, and story memory. No other significant associations with PAE noted Frequency of binge drinking events in the first trimester of pregnancy predicted poorer outcomes on WRAML Design Memory (β=-1.51); verbal learning (β=-1.55); and story memory (β=-1.25).	Average alcohol exposure was negatively associated with WRAML Design memory, verbal learning and story memory.
Motor Skills	Maternal Health Practices	(Richardson et al., 2002)	Grooved Pegboard	10 (*n*=593)	Average number of drinks per day during pregnancy, split by trimester	First trimester alcohol consumption predicted performance on WRAML Design memory, verbal learning, and story memory. No other significant associations with PAE noted	No associations were reported between average alcohol exposure per day and fine motor skills, as measured by the grooved pegboard tests.

Chapter 3 Qualitative Studies

Preface

The introduction to this thesis (Chapter 1) highlighted that there is an opportunity for developing a new screening tool for children who display cognitive and behavioural functions consistent with children with FASD. The results of the scoping review in Chapter 2 suggest that while children with FASD, and PAE, display a range of cognitive and behavioural impairments, there is evidence for broadly consistent differences in several domains of functioning. While information gathered from the literature review is valuable in informing the initial development of a screening tool, additional research with caregivers and clinicians of children with FASD is required to identify the full spectrum of cognitive and behavioural functioning among children with FASD. Qualitative research is important in measure development to ensure items are representative of, and relevant to the construct of interest (Haynes, Richard, & Kubany, 1995).

As such, the second phase of this PhD research was to conduct qualitative research to explore in-depth, caregivers' and clinicians' perspectives of the cognitive and behavioural functioning of children with FASD. In all, 14 caregivers, and five clinicians participated in semi structured telephone interviews. Content analysis was undertaken to identify themes from the interviews to strengthen and add to aspects of cognitive and behavioural functioning displayed by children with FASD that were identified in Chapter 2 (Scoping Review) of this thesis. These interviews provide insight into the caregivers' and clinicians' perspectives on the common challenges experienced by children with FASD.

Structure of Chapter 3

Chapter 3 is presented in two parts. The first part, Chapter 3A presents a submitted manuscript of interviews undertaken with caregivers of children with FASD. This qualitative

study has been submitted for publication to the *Research in Developmental Disabilities* journal, received feedback and has been resubmitted with suggested amendments. Due to length restrictions on the manuscript, additional quotes from caregivers which illustrate the coding and content analysis could not be incorporated. These are presented in Appendix A. Based on initial reviewer feedback during the peer review process, information pertaining to the clinician's interviews that were previously included in the manuscript were removed from the publication due to a lack of data saturation due to the small number of participants recruited. Given the dearth of information available in the literature regarding clinician's perspectives of the cognitive and behavioural characteristics of children with FASD, it was thought important to include this information as part of the PhD. As such, the findings related to the clinician's interviews are presented in Chapter 3B. Further, in Chapter 3B, the findings of the interviews with clinicians will be compared and contrasted with the findings from caregiver interviews.

Chapter 3A Caregivers Interviews

A Qualitative Examination of the Cognitive and Behavioural Challenges Experienced by Children with Fetal Alcohol Spectrum Disorder

Accepted for publication in *Research in Developmental Disabilities*

Abstract

Introduction: At present it is unclear whether there is a consistent behavioural phenotype for children with Fetal Alcohol Spectrum Disorder (FASD) that can support screening efforts. There has been a dearth of qualitative studies exploring the behavioural phenotype from the perspective of caregivers raising children with FASD. The current study explores the cognitive and behavioural difficulties and impairments experienced by children with FASD aged between four and 12 years from the perspective of caregivers.

Methods: Fourteen caregivers of children with FASD participated in telephone interviews. Caregivers were recruited until data saturation occurred. Thematic analysis was undertaken on the transcribed interviews, using NVivo 12.

Results: Three over-arching themes were identified that consisted of subthemes 1) Self-regulation; behavioural, emotional, and attention; 2) Cognitive abilities; academic abilities and learning and memory; and 3) Adaptive functioning; social skills, communication and language skills, motor skills, and sleep concerns. Multiple subthemes were consistently identified across participants. A further two cross-cutting themes were identified; children behaving young for their age, and inconsistency in behaviour and strategies.

Discussion: Despite the lack of a consistent behavioural phenotype for FASD, the findings suggest consistency between caregivers in their reports of the difficulties experienced by children with FASD. The implications for early identification and screening tool development are discussed.

Keywords: Fetal Alcohol Spectrum Disorder (FASD), qualitative research, children.

What does this paper add?

Many quantitative studies have been conducted examining cognitive and behavioural outcomes of Prenatal Alcohol Exposure (PAE), and by extension, amongst children and adolescents with Fetal Alcohol Spectrum Disorder (FASD). However, there have been few qualitative studies of these outcomes and fewer still investigating young children. Qualitative methodologies exploring the nuances and full spectrum of difficulties experienced by children living with FASD can contribute to improved tools for early identification, as well as provide insight into the support needs of children and families. This study contributes to the literature by exploring caregiver perspectives on the difficulties experienced by children living with FASD.

Highlights

- There are few qualitative studies of impairments among children with Fetal Alcohol Spectrum Disorder.
- We conducted semi-structured interviews with caregivers of children with Fetal Alcohol Spectrum Disorder.
- Consistent themes in cognitive and behavioural difficulties were identified.
- Implications for screening and early identification are discussed.

Introduction

Children prenatally exposed to alcohol can experience a range of cognitive and behavioural impairments. Fetal Alcohol Spectrum Disorder (FASD) is the umbrella term for a range of diagnoses reflecting the outcomes of Prenatal Alcohol Exposure (PAE). Several countries (e.g., Australia and Canada) have adopted FASD as the diagnostic term applied to individuals who demonstrate cognitive and behavioural impairments, across at least three cognitive and behavioural domains, following PAE (Bower & Elliott, 2016; Cook et al., 2016). FASD prevalence has been estimated at between 1% and 5% of school-aged children in the USA (May et al., 2018) and Canada (Popova et al., 2019). Recent UK research estimates up to 17% of children may have FASD (McQuire et al., 2019). Currently, no accurate population prevalence rates for Australia are available, with passive surveillance rates thought to underestimate FASD prevalence (Reid, 2018).

Children with FASD may go unrecognised or misdiagnosed and consequently early intervention opportunities can be missed. For example, in a study of 547 children who had previously undergone multi-disciplinary assessments, when re-examined, 156 children met FASD diagnosis criteria, and of these, FASD had been missed in 80% of cases (Chasnoff, Wells, & King, 2015). Many children with FASD do not display the dysmorphological features typically associated with FASD (i.e., short palpebral fissures, thin upper lip, and an indistinct philtrum) and neuropsychological impairment may be independent of the presence of physical features (Mattson, Riley, Gramling, Delis, & Jones, 1998). Furthermore, children who do not show FASD facial features are often diagnosed later and experience poorer long-term outcomes than children who do present with the physical features of FASD, with the latter finding attributed to a delay in receiving support (Fagerlund, Autti-Rämö, Hoyme, Mattson, & Korkman,

2011; Streissguth et al., 2004). Early identification of children at-risk of FASD is vital to ensure children receive appropriate support and early intervention. A missed or misdiagnosis may result in caregivers receiving general parenting advice, rather than therapeutic supports tailored for children with FASD (Chamberlain, Reid, Warner, Shelton, & Dawe, 2017).

It has been suggested that a behavioural phenotype, defined as the characteristic pattern of motor, cognitive, linguistic, and social observations that are consistently associated with a biological disorder (O'Brien and Yule, as cited in Kodituwakku (2007)) could aid in the identification and diagnosis of FASD. Despite decades of research, a FASD behavioural phenotype has not yet been identified, owing in part to the complex relationship between PAE and subsequent outcomes. While a specific behavioural phenotype that allows for diagnosis in itself may be unlikely (Lange, Shield, Rehm, Anagnostou, & Popova, 2019), identifying the common challenges experienced by children with FASD that are relevant to caregivers may be informative for screening. Recognising such challenges could lead families and clinicians to more easily identify the need for further assessment and consider the potential for prenatal exposures among differential diagnosis.

Qualitative research offers an avenue to explore the cognitive and behavioural functioning of children with FASD from the perspectives of caregivers. Furthermore, qualitative research complements the extensive quantitative research of children's cognitive and behavioural functioning and allows insight into aspects of functioning that may not be identified using quantitative measures. Caregivers have extensive experience in supporting their children and can therefore speak to the range of cognitive and behavioural difficulties displayed by their children. While clinical assessments may involve caregiver-reports of children's behaviour, caregivers also understand situation-specific triggers and observe behaviours not appearing on standardised

clinical checklists or measures. Although previous qualitative research has explored various aspects of raising a child with FASD (Domeij et al., 2018), qualitative literature examining caregiver perspectives on the cognitive and behavioural profile of children with FASD is scarce. Such information is critical in developing valid and reliable tools to assist with early screening of FASD (Streiner, Norman, & Cairney, 2015).

While a number of qualitative studies have been conducted examining the cognitive and behavioural impairments of individuals with FASD from caregivers' perspectives, these have typically included combined samples of children, adolescents, and young adults with FASD, ranging from five to 30 years old (Salmon, 2008; Sanders & Buck, 2010; Whitehurst, 2012). To date, one qualitative study has been conducted examining behavioural and cognitive impairments of children (up to 18 years old) with FASD from caregiver perspectives. Children with Fetal Alcohol Syndrome (FAS; i.e., those children displaying the three sentinel facial features) aged between two and 16 were described by caregivers as being unable to retain information or understand single-step instructions, lacking an understanding of the consequences of their actions, lacking fear or judgement, being hyperactive, aggressive, destructive, and having a high pain tolerance (Gardner, 2000). Qualitative studies amongst the combined samples of children, adolescents, and young adults with FASD identified similar themes including difficulty understanding consequences, lack of judgement, hyperactivity, tantrums, aggressive behaviour, and social immaturity (Salmon, 2008; Sanders & Buck, 2010; Whitehurst, 2012).

In the current study, we sought to explore caregiver perspectives of the cognitive and behavioural functioning of four to 12-year-old children with FASD. Domains of impairment, such as executive functioning, become more evident as children move into school, and demands on their cognitive capacities increase (Campbell, Halperin, & Sonuga-Barke, 2014). Identifying

the common challenges experienced by school-aged children with FASD may assist in the development of screening tools for children in this age range. Moreover, understanding these challenges from the perspective of caregivers may help to develop educational materials that support caregivers to understand when seeking assessment and support may be appropriate.

Methods

Participants

Participants were Australian caregivers raising children with FASD. Caregivers were eligible to participate if they were primary caregivers (biological parents, kinship carers, foster carers, adoptive parents or legal guardians) of a child between the ages of four and 12 years, with a FASD diagnosis or provisional diagnosis. Given the relatively recent introduction of the Australian FASD diagnostic guidelines (Bower & Elliott, 2016), participants were eligible regardless of the diagnostic classification under the broader FASD umbrella their child received. Verification of the diagnosis through medical records or other sources was not sought due to the complexity of this process.

Thirty-three caregivers contacted the research team, with 14[9] (42.4%) participating in interviews. Reasons for non-participation included non-responses at follow up, or individuals based outside Australia. Information regarding numbers of potential participants reached through social media networks and mailing lists was not available, thus an overall response rate cannot be calculated. Table 1 summarises demographic characteristics of participating families. Caregiver participants were all non-biological caregivers of children with FASD. Participants came from four Australian states. Most participants were female (93.3%). The 14 caregivers were raising 17

[9] The total number of interviewees was 15 due to both foster mother and father from one family participating. For ease of interpretation, the authors refer to 14 participants throughout the paper.

children with either diagnosed, or provisionally diagnosed FASD. Children had a mean age of 8.06 years old (*SD*=2.28 years). Where caregivers had additional children who they believed to have FASD but had not been diagnosed, they were asked to speak only about the child/ren who had a confirmed or provisional diagnosis. Caregivers varied in the length of time children had been in their care, with some caregivers having children from birth, whereas others had children placed in their care at ages four and five years (see Table 1). Several caregivers discussed children experiencing child maltreatment prior to being placed in their current placement.

(Insert table 1 about here)

Procedure

Ethical approval was obtained from the University of South Australia Human Research Ethics Committee (Protocol number: 0000036058). Ethics approvals were limited to Australian participants, hence potential overseas participants were excluded. A purposive recruitment method was employed for recruitment, with a study invitation distributed through mailing lists and social media accounts of two Australian organisations supporting families living with FASD. The study invitation provided the rationale for the research, the first author's contact details and a link to an online questionnaire where potential participants could leave contact details for follow up.

Telephone interviews were used given the wide Australian geographical distribution of families affected by FASD. This is consistent with previous studies (Breen & Burns, 2012). Participants were not known to the interviewer before they consented to participate. Rapport was established with participants through self-disclosure of interest in the research area, displaying an interest in participants experience and regarding families as experts (Cridland, Jones, Caputi, & Magee, 2015). Caregivers who indicated an interest in participating were sent physical copies of

the information and consent forms to review. Prior to the interview commencing, the purpose of the research was explained, and the consent form was read to participants by the interviewer and participants provided verbal consent. The telephone interviews were conducted in a private office. Interviews lasted on average 53 minutes (range = 31-107 minutes). Participants were sent copies of their interview transcript and a reply-paid envelope and invited to provide additional comments or clarification, should they wish to. No additional comments were received from participants. Caregivers received an AU $20 gift card for their time.

A semi-structured interview guide was developed (see Supplementary File 1). Caregivers were asked about the functioning of their children with FASD in their day to day lives, including at home and school, their relationships with others, strengths their children displayed, and the cognitive or behavioural symptoms that caregivers first noted. The interview guide was trialled with one family, and revised. This interview data was not included in analysis. As interviews had the potential to be distressing, participants were provided with the details of organisations that could provide support should they experience distress. Participants were also encouraged to have a support person with them if they felt it necessary.

Data Management and Analysis

Interviews were audio-recorded with participants' consent and transcribed verbatim, with potentially identifying information removed. Field notes were made during interviews and these noted initial ideas for transcript analysis. Transcripts and hand-written notes taken during interviews were imported into NVivo 12 Pro (QRS International, 2018) for analysis. A qualitative judgement on data saturation was made based on the fourteenth interview, that no new themes were being identified during the interviews (Brod, Tesler, & Christensen, 2009). Thematic analysis (Braun & Clarke, 2006) was used to identify key themes regarding cognitive

and behavioural challenges experienced by children with FASD. An inductive approach to data analysis was adopted and analysis was conducted following the steps of thematic analysis (Braun & Clarke, 2006).

1. The first author transcribed all interviews, checked transcriptions for accuracy, and read and re-read each transcript.
2. Initial codes were generated and discussed in regular meetings between the first author and the research team.
3. Codes were collated into potential sub-themes and major themes. Existing frameworks were drawn upon to assist the interpretation of the major-themes and sub-themes that emerged from participants' responses. Existing frameworks included the Australian FASD diagnostic guidelines (Bower & Elliott, 2016), Neurodevelopmental Disorder associated with PAE (American Psychiatric Association, 2013), and reviews of the cognitive and behavioural functioning of children with FASD (Kodituwakku & Kodituwakku, 2014).
4. The themes were reviewed and revised and checked against both the coded extracts and the original data set. For the purposes of inter-rater reliability, a random selection of five transcripts (36%) were reviewed against the coding by a second coder from the research team.
5. The codes were further refined, named, and defined.

All interviews and initial analysis were conducted by the first author (SM), a male psychology doctoral candidate and research assistant with previous experience in conducting qualitative research with vulnerable participants. The research team included researchers with experience in child development, addiction and substance use, trauma, and FASD. The research

team's knowledge of FASD and child development informed the qualitative analysis. To reduce any potential bias associated with a sole researcher conducting all data collection and coding, regular meetings were held between the first author and the research team throughout the data analysis. Further, for inter-rater reliability a random selection of transcripts was reviewed by the second and fourth authors against the coding frame to ensure codes accurately reflected interview content, and no potential themes were missed.

Results

Caregivers described difficulties experienced by their children compared to their peers and other children of a similar age. Several caregivers drew on their experience with their own, often similarly aged, biological children. While some of the behaviours may be developmentally appropriate, children appeared to have not "grown out" of these stages. Three over-arching themes were identified which included major themes and subthemes. The over-arching and major themes were: (1) Self-regulation, including behavioural, emotional, and attention regulation; (2) Cognitive abilities, including learning and memory and academic abilities; and (3) Adaptive functioning, including communication and language skills, social skills, motor skills, and sleep concerns. A further two themes were identified as recurring across the three over-arching themes; Young for age, and Inconsistency in behaviour. Figure 1 shows the over-arching themes, major themes, and their sub-themes. Table 2 provides exemplars and the numbers of caregivers describing experiences consistent with each theme.

(Insert Figure 1 about here)

(Insert Table 2 about here)

Self-regulation

Behavioural regulation

Caregivers raised concerns about children's abilities to regulate behaviour. Four sub-themes were identified; hyperactivity, impulsivity, risk-taking behaviours, and taking things which don't belong to them. Caregivers described children as "being unable to sit still, constantly moving or making noise". Impulsivity often manifested as risk-taking behaviours, for example running into traffic, or jumping into bodies of water fully clothed, with potentially harmful consequences for their children. Impulsivity was compounded by difficulties understanding or anticipating the consequences of actions. Caregivers described a tendency for children to take things which did not belong to them, for example items from the supermarket or hardware store, other children's school lunches, and caregiver's jewellery.

Emotional regulation

Caregivers raised concerns about children's ability to regulate their emotions. Two subthemes were identified; tantrums or meltdowns and managing transitions. Children were described by caregivers as being "prone to tantrums", which were often over seemingly trivial things, and longer and more intense than caregivers expected from children their age. One caregiver described a twelve-hour tantrum after asking their child to put on a different pair of shoes. Caregivers also described their children as having difficulty in calming down once a tantrum or meltdown had started. The explosive and variable nature of children's emotional reactions were a cause of significant caregiver stress. A common trigger to meltdowns was changes to routines and transitions between activities. Caregivers described needing a "long lead-time" with frequent reminders about an impending transition. In situations where caregivers had to rush their children between activities, behavioural regulation issues would be exacerbated,

as one caregiver described “the brakes would go on”, and children could struggle to engage in the activity.

Attention regulation

Attention regulation was comprised of two sub-themes; being “easily distracted” and needing external support to help children to stay focused. Caregivers often linked difficulties with attention to difficulties with short-term or working memory, as one caregiver described their 11-year-old child as “getting distracted and immediately forgetting an instruction”. Caregivers noted that children often “needed constant reminding”, or supervision to stay focused on a task in the classroom.

Cognitive abilities

Learning and memory

Caregivers described four sub-themes relating to learning and memory; working memory, difficulty understanding consequences, forgetting previously learnt information and needing repeated exposure for information retention. Working memory difficulties primarily manifested as difficulties completing multi-step instructions and were closely linked with descriptions of attention difficulties. One caregiver described her 11-year-old daughter as “getting distracted and forgetting instructions as soon as she turned around to perform whatever it is”. Further, caregivers reported their children as not “getting it at all that there were consequences of their actions” and did not learn from their mistakes. Difficulties understanding consequences impacted upon caregivers’ strategies for discipline, with strategies like time-out described as ineffective as “children would forget why they were in time-out”. Caregivers described a pattern of forgetting things from one day to the next, wherein caregivers reported working with children on skills such

as spelling lists or telling the time until they displayed proficiency, but children were unable to retain that knowledge the following day.

Academic abilities

Caregivers described three sub-themes relating to academic abilities; general school performance, maths, and literacy skills. Caregivers reported that children had significant difficulties with maths, particularly with increasingly complex or abstract problems. Observations by caregivers regarding problems with literacy were mixed; and some caregivers reported difficulties with reading and comprehension, whereas others identified literacy as a relative strength and reported their children to enjoy reading. Difficulties with writing were also reported and these were often closely related to motor skills difficulties (e.g., difficulties holding pens, pencils, and crayons). A small number of caregivers characterised their children as appearing more capable when they had prompts or could copy content, whereas they had greater difficulties "getting the words from their head on to the page". Six children were reported to be completing work at grade levels below that of same-age peers. Children's abilities in the classroom were often impacted by difficulties in other domains, such as sustaining attention. Caregivers noted children were more able to engage when classroom accommodations such as one-on-one attention or visual aids were made available.

Adaptive functioning

Communication and language skills

Three sub-themes relating to communication and language skills were identified: receptive language difficulties, questioning whether children were wilfully defiant or not understanding, and lying or confabulation. Caregivers described children as not understanding what was said to or asked of them. For many caregivers, difficulties described centred around

following lists of instructions, which also related to working memory and attention impairments. Caregivers interpreted difficulties following instructions as “not listening”; and questioned whether children were being “deliberately defiant, or not understanding what was being said”. Caregivers attributed this to language processing difficulties, and not comprehending instructions. Further, some caregivers noted their children displayed delays in auditory processing and would wait 10 to 15 seconds before repeating an instruction, particularly if the children were in a heightened state.

Caregivers described children as often appearing to lie or confabulate, although caregivers questioned whether this was intentional behaviour to escape trouble or reflecting a genuine failure to recall events. Caregivers found this challenging as children appeared sincere in their belief that they were telling the truth and could become upset if challenged. Confabulation was seen as reflecting a failure of memory, with children “filling in the blanks” and being suggestible about what had happened, or “confusing dreams with memories” Confabulation descriptions may reflect caregivers reframing their child’s behaviour in light of their understanding of the underlying impairments associated with FASD.

Social skills

Four sub-themes relating to social skills were identified; difficulties making and keeping friends, understanding social cues, having a friendly nature, and displaying a preference for younger peers. Caregivers described children as friendly but struggling to engage appropriately and to make and keep friends. Further, children were described as having difficulty understanding social cues and perspective-taking. For example, caregivers reported children having an apparent disregard for others’ personal space or struggling to understand the rules in contact sports. Children were unaware of the impact of their behaviours on others, with

caregivers describing children as not realising when their behaviour was annoying others. Children with FASD were reported to often be excluded by peers who did not understand their behaviour. Children's difficulties with communication including difficulties understanding humour or non-literal language, also impacted upon their relationships with their peers. Several caregivers raised concerns about their children being indiscriminately friendly towards strangers.

Motor skills

Some caregivers raised concerns about their children's gross motor skills, regularly describing them as clumsy. Caregivers were able to focus on and work with children to improve gross motor and coordination skills when running, bike riding, balancing, and swimming. In contrast, other caregivers described their children as having a relative strength in their gross motor skills. For these children, difficulties with hyperactivity, impulsivity, and understanding the consequences of actions became a larger concern, as children were more capable of placing themselves into potentially dangerous situations (e.g., six and seven year old children climbing on to the roof). Fine motor skills were often described as impaired, with children described as having difficulties with handwriting and using cutlery. Caregivers described their children as struggling at school due to their handwriting, and for their handwriting to look more like that of chronologically younger children.

Sleep concerns

Caregivers described a range of challenging sleep-related behaviours including children being very light sleepers, taking a long time to get to sleep, frequent night-waking, having nightmares, or short sleep times. Moreover, six children were reported to have been prescribed both sleeping medication and melatonin. Disrupted sleep placed additional strain on caregivers, the wider family unit, and the child the following day. Caregivers were less able to function, had

less energy for their other children, and the children with FASD had greater difficulty with behavioural regulation following a poor night's sleep.

Cross-cutting themes

Young for their age.

Caregivers described children as "immature for their age" or "young-minded". This theme was identified as a crosscutting theme as it was present across several overarching themes and subthemes such as behavioural regulation, emotional regulation, academic ability, and social skills. The theme Young for their Age was most evident in relation to the themes of children's emotional regulation with caregivers describing children as behaving or responding to their peers like a chronologically younger child. In discussing relationships with peers, caregivers reported children with FASD often preferred the company of younger peers, and the play skills demonstrated by children with FASD were behind that of their peers. Lastly, children with FASD were often reported to be assigned schoolwork below that of their chronological age showing the cross-cutting relationship with the theme of academic abilities.

Inconsistency in behaviour and strategies.

Caregivers described patterns of children behaving inconsistently which was reported to occur across the three over-arching themes. Some children were described as inconsistent in their emotional and behavioural regulation, whereas other children were inconsistent with respect to academic abilities. One caregiver reported teachers attempting to "squeeze in as much information as possible" on days where her eight-year-old child's memory appeared better. Caregivers found this inconsistency challenging to respond to, as they were not certain about what behaviour they would experience from day to day and reported finding themselves anxious in expecting behaviour change. Further, caregivers explained that behaviour management

strategies which worked some days, may not work on others and their strategies had to be adaptable.

What Cognitive and Behavioural Symptoms Did Caregivers First Notice in Their Children?

Caregivers were asked whether there were behaviours or cognitive challenges displayed by their children that led them to suspect they may have FASD. Behavioural regulation difficulties (particularly hyperactivity), and emotional regulation difficulties were common concerns identified by caregivers as the first potential signs of FASD. Several caregivers highlighted preschool and entry into school as times when these behaviours became more pronounced. Additional concerns commonly identified by caregivers as early warning signs including difficulties with executive functioning and working memory, lack of inhibition, classroom learning difficulties, and challenges learning adaptive skills such as dressing independently. Caregivers' early concerns about learning difficulties often centred around children not learning from their mistakes or forgetting previously learnt information. A small number of carers initially attributed the behaviours that they considered challenging to placement changes before children came to live in their current home, being in foster care or having past trauma experiences.

Delays in children's development and milestones (e.g., speech, walking, self-care behaviours) were another initial cause of concern for caregivers. Caregivers reported health and development concerns with difficult feeding or restrictive eating, children often getting sick, issues in getting children to sleep, and impaired motor skills. A small number of caregivers

reported initial concerns regarding shaking, trembling, or rocking in very young children[10]. Delays in walking, and toileting were reported, albeit not universally.

Strengths

Caregivers were asked to describe the strengths displayed by their children. A range of strengths were described, with no clear patterns emerging. As described above, several caregivers identified motor skills, and children's interest in sports and physical activity as a strength. One caregiver was able to adapt an interest in play sports to supporting teaching maths skills such as addition and multiplication. Other caregivers commented on stronger memory for topics that were of interest to their children, such as remembering and avoiding duplication in a collection of toy cars. Caregivers also described their children as being very friendly and loving and being capable of being very engaging in conversation.

Discussion

The aim of this study was to explore the cognitive and behavioural functioning of children aged four to 12 years old with FASD, from the perspective of their caregivers. We sought to identify common challenges that may inform the development of a screening tool. Three overarching themes were identified: 1) Self-regulation, 2) Cognitive ability, and 3) Adaptive functioning. Given the complex biological mechanisms by which alcohol affects development (Sulik, 2014), variability in patterns of exposure (Flak et al., 2014), and moderating factors (May et al., 2016; May et al., 2014), the varied presentation of children with FASD is unsurprising. While there was some variation in the behavioural concerns identified by

[10] This was only identified by a small number of participants and was not identified as a theme owing to the small number who described such behaviour. It should be noted, however, that the caregivers who did identify this theme had children in their care since birth and such behaviours may have occurred for other children prior to their current placements.

caregivers of children with FASD, caregivers described a consistent presentation of cognitive and behavioural difficulties across these three domains. Further, it is important to note that there was an inter-relationship between the themes and difficulties in some areas that may influence performance in others. For example, lying or confabulation may reflect memory difficulties, and difficulties in behaviour regulation may impact upon social skills and peer rejection. These insights may be used to inform the development of observer-completed screening tools to improve the early identification of children living with FASD. While existing screening tools have focused on externalising behaviours (e.g., the Neurobehavioural Screening Tool; see Lange, Rovet, Rehm, and Popova (2017) for review), the current study results suggest a broader view of the cognitive and behavioural difficulties. Further, findings of this study were consistent with previous qualitative research which found children, adolescents, and young adults with FASD to have difficulties with understanding consequences of behaviour, lacking judgement, forgetting previously learnt concepts, hyperactivity, and difficulties engaging with peers (Gardner 2000; Salmon, 2008; Sanders & Buck, 2010; Whitehurst, 2012). In addition to informing the development of screening tools, these results highlight key areas where targeted supports for children with FASD may be required.

The first over-arching theme related to difficulties in self-regulation, including regulation of behaviour, emotion, and attention. Caregiver descriptions of this theme centred around hyperactivity, impulsivity, difficulties staying focused on a task, and emotional regulation. The pronounced difficulties reported for this theme align with findings from a recent review of self-regulation amongst children with PAE (Reid & Petrenko, 2018). Reid and Petrenko (2018) examine the developmental hierarchy of the self-regulatory skills, whereby basic self-regulatory skills, such as physiological regulation, are essential for the development of higher order

regulatory skills such as behavioural regulation and executive functioning. In turn, executive functions are considered to be a necessary, but not sufficient component for overall self-regulation (Hofmann, Schmeichel, & Baddeley, 2012), and previous work by Kable and colleagues has separated executive function impairments from impairments in self-regulation among children with ND-PAE (Kable et al., 2016). While Kable and colleagues describe two components of self-regulation (mood/arousal and attention/impulse control), with behavioural dysregulation described in both areas, this theme emerged as a distinct, but overlapping area in the current research. Caregivers' descriptions of children's behaviour focused on the self-regulatory aspects of executive functioning, such as difficulties with impulse control and behavioural regulation. The cognitive aspects of executive functioning (e.g., poor planning, difficulty changing strategies, problem solving strategies) were less frequently discussed and while children may experience difficulties in these areas, the self-regulatory difficulties may be more salient in everyday life. Previous research has shown children with FASD have significant impairment in executive functioning (Kingdon, Cardoso, & McGrath, 2015). Given that the brain regions associated with executive functioning may be particularly sensitive to PAE effects (Gautam, Nuñez, Narr, Kan, & Sowell, 2014), regulatory difficulties may be one of the FASD "hallmark" features. Importantly, self-regulation appears amenable to intervention among children with FASD. Children with FASD who have completed the *Alert* (Williams & Shellenberger, 1996) self-regulation intervention have shown improvements in parent- and teacher- ratings of behaviour and executive function, as well as objectively measured executive functioning (Reid et al., 2015).

The second over-arching theme related to cognitive ability. Sub themes included learning and memory, and academic abilities. Caregivers described children as having poor short-term or

working memory, needing repeated exposure to learn new information, appearing to forget previously learnt information, and experiencing difficulties understanding consequences of their actions. Working memory is typically included in definitions of executive function, difficulties with working memory were included under the memory themes as this reflected how caregivers spoke about the memory difficulties displayed by their children. Memory difficulties have been commonly reported among children with FASD (du Plooy, Malcolm-Smith, Adnams, Stein, & Donald, 2016). Caregivers' reports of academic difficulty were intertwined with behavioural and attention regulation difficulties, memory difficulties, and language difficulties. For children with FASD experiencing difficulties with behavioural, attention and emotional regulation, they may therefore experience significant difficulties with classroom engagement. These findings were consistent with previous research on academic abilities, in particular with regards to the mathematics difficulties experienced by children with FASD (Rasmussen & Bisanz, 2009) and the relationships between difficulties in other domains impacting negatively upon classroom behaviour and performance (Millians, 2015). Hyperactivity, and difficulties inhibiting behaviour, and staying focused- particularly when a task was challenging- were all described by caregivers as contributing to off-task behaviour in the classroom. These findings were consistent with previous research suggesting that children with FASD demonstrate performance decrements with increasing task complexity (Kodituwakku & Kodituwakku, 2014).

The third over-arching theme related to adaptive functioning. Limitations described by caregivers aligned with previous research that found significant difficulties with communication and language skills (Wyper & Rasmussen, 2011) social skills (Kully-Martens, Denys, Treit, Tamana, & Rasmussen, 2012), and motor skills (Lucas et al., 2014) among children with FASD and heavy PAE. Previous research has highlighted the importance of behavioural regulation as an

important predictor of social skill abilities, while another study found that children with FASD have greater difficulties understanding their emotions and the emotions of others (Petrenko, Pandolfino, Quamma, & Carmichael Olson, 2017). These findings are consistent with the current study and demonstrate the pervasive and interconnected functional impairments experienced by children with FASD.

Caregivers in this study described difficulties relating to sleep including children being very light sleepers, taking a long time to get to sleep, frequent night-waking, having nightmares, or short sleep times. While sleep concerns may not be part of the current FASD diagnostic assessment, the assessment of sleep concerns is important for intervention planning (Hanlon-Dearman, Chen, & Olson, 2017). Previous research has suggested that sleep concerns among children with FASD are often missed by health care providers (Ipsiroglu, McKellin, Carey, & Loock, 2013). Emerging evidence suggests that parent-delivered behavioural sleep interventions may be used for children with a range of neurodevelopmental disorders (Rigney et al., 2018). Given sleep quality impacts cognitive and behavioural functioning of children and can cause significant impacts on the wider family (Hanlon-Dearman et al., 2017; Inkelis & Thomas, 2018), trialling these interventions with children with FASD is an important future research direction.

Another important finding from the current study was that caregivers consistently described their children as acting younger than their chronological age, as well as inconsistency in their child's behaviour and the effectiveness of strategies used to manage their child's behaviour. Previous research has similarly described children with FASD as showing a pattern of dysmaturity- that is functioning socially, emotionally and cognitively at a level below their current age (Malbin, 2004). This theme was identified as crosscutting, as it was raised in relation to several of the other themes and sub-themes identified. Caregivers reported their children's

behaviour being inconsistent over time and noted that it was often difficult to predict. This finding was consistent with previous research finding children with FASD to display greater variability in performance on neuropsychological tasks over time (Ali, Kerns, Mulligan, Olson, & Astley, 2018), as well as greater variability in their classroom behaviour across days (Kjellmer & Olswang, 2013). This indicates a need for intervention strategies to be targeted at a child's developmental age rather than their chronological age. A number of interventions have been developed for children with FASD which adapt their content to suit the developmental age of children including Families Moving Forward (Petrenko, Pandolfino, & Robinson, 2017), Maths Interactive Learning Experience (Kully-Martens et al., 2018), and the GoFAR program (Coles, Kable, Taddeo, & Strickland, 2018). Further, resources for caregivers raising children with FASD stress the need to understand children's developmental, rather than chronological age. Moreover, it suggests a need for repeated assessment over time to understand the full range of a child's cognitive and behavioural difficulties. Previous research has suggested that developing an understanding of FASD, the brain-based nature of the behavioural problems, and reason why traditional strategies may not be appropriate for their children is critical for carers of children with FASD to adapt their strategies accordingly (Coons, Watson, Schinke, & Yantzi, 2016).

The early identification of children who may have FASD is important to help ameliorate long term difficulties associated with the disorder. For many caregivers in the current study, hyperactivity and emotional regulation were early indicators of the need for investigation with several children receiving an ADHD diagnosis prior to their FASD diagnosis. This finding is unsurprising, given the high rates of co-morbidity between FASD and ADHD (Chasnoff et al., 2015). However, it indicates the need for comprehensive assessment of prenatal exposures when undertaking diagnostic testing for neurodevelopmental disorders. Caregivers also described

concerns with learning and memory as early warning signs. It was also noted that problems became apparent after children started formal education. These findings suggest that routine screening points – such as on-entry school assessments – may represent an important opportunity to screen and refer children at risk of developmental disorders, including FASD. Ideally, it would be best to identify children with FASD as close to birth as possible and to provide a range of early interventions as necessary. However early childhood is often a difficult time to identify children with FASD especially among children who may not display the physical features associated with FASD, or who demonstrate less severe delays. While children who display the facial features or microcephaly may be readily identifiable, the accurate assessment of cognitive and behavioural impairment amongst younger children represents a barrier to early diagnosis (Cook et al., 2015; McLachlan et al., 2015). Novel approaches to identifying children such as eye-tracking (Zhang et al., 2019), or the cardiac orientating response (Mesa et al., 2017) may have potential to overcome the difficulties in behavioural assessment with younger children (Finlay‐Jones et al., 2019).

An inductive approach was adopted in analysing the participant data, and while the major themes and sub-themes were not predefined, existing frameworks were used to inform the organisation of major themes into over-arching themes. The emergent major themes (See Fig. 1) mapped closely to ND-PAE criteria (American Psychiatric Association, 2013). The ND-PAE core symptoms include neurocognitive impairment, impairment in self-regulation, and deficits in adaptive functioning (Kable et al., 2016). The overlap noted between the common difficulties discussed by caregivers in this study and the ND-PAE core symptoms suggest that screening tools reflecting these domains may have utility in identifying children who display cognitive and behavioural impairments consistent with FASD.

Several limitations need to be considered in relation to this study. Caregivers self-reported their child as having a FASD diagnosis and additional sources of information were not sought to confirm the diagnosis. Further, several children were identified as having a provisional FASD diagnosis. Provisional diagnoses reflected children too young to receive full neurocognitive testing or children who are confirmed to have had PAE but who were awaiting further assessments. There is the potential that upon further testing the child/ren, will receive a diagnosis other than FASD, or may receive no diagnosis. Lastly, reflecting the recent publication of the Australian Guide to the Diagnosis of FASD (Bower & Elliott, 2016), there may be heterogeneity in the criteria against which children were assessed at the time of their diagnosis.

There is a potential that the sample is biased as recruitment occurred through support organisations. None of the caregivers were birth parents of the children and several caregivers identified their children as having a history of child maltreatment. Exposure to adverse childhood events including abuse, neglect or multiple home placement may compound the cognitive and behavioural difficulties experienced by children with FASD (Price, Cook, Norgate, & Mukherjee, 2017). Children varied in the time they were removed from their birth parents; ranging from removal at birth, to being placed in care at age five. Given there were differences in the developmental stages that participants in the current study had been exposed to, there is likely variation in their perspectives of early warning signs of the disorder. A subset of participants were able to identify signs evident from birth (e.g., difficulty feeding). Future research could seek to further explore early warning behaviour patterns observed by parents and carers among younger children, and to conduct qualitative research with clinicians and specialists who work with this group as they would be able to note consistent impairments across a broader range of children with FASD.

Conclusion

Caregivers described a broad, but consistent set of challenges experienced by children with FASD. These findings identify a consistent presentation observed in a sample of caregivers that can assist with the development of FASD screening tools. Understanding common challenges experienced by caregivers of children with FASD and recognising the common cognitive and behavioural functioning difficulties displayed by children is an important step in developing screening tools which may support earlier identification and intervention strategies.

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Conflict of interest statement

The authors have no conflicts of interest to report.

Tables

Table 1

Family and Child Demographics

			Child			
ID	Gender	Relationship	Age/s	Gender/s	Diagnoses	Child age (years) when placed in the current placement
P1	F	Kinship carer	10	M	pFAS	Since birth
P2	F	Foster carer	10, 8	M, M	ARND, FASD	Youngest was 2.5 years
P3	F	Foster carer	10	M	FAS	Since before preschool
P4	F	Kinship carer	8	F	FAS	14 months
P5	F	Legal guardian	11	F	FASD	First placed at 6 years of age for 12 months and again following reunification attempt.
P6	F	Foster carer	9, 6	M, M	SE:AE, Provisional	Eldest placed at 5 weeks, Youngest since birth
P7	F	Adoptive parent	12	M	FASD	At least 18 months
P8	F	Legal guardian (Aunt-in-law)	9	F	pFAS	Since 5 years old
P9	M	Kinship carer	10, 8	F, M	FASD	Since approximately 2 years old
P10	F	Foster carer	8	M	FASD	Since 2 weeks old
P11	F	Kinship carer	5	F	Provisional	Since birth
P12	F	Kinship carer	7, 6	F, F	FASD	Eldest placed at 3.5 years, youngest at 2.5 years
P13[1]	F, M	Foster carer	6	F	Provisional	Placed at 2.25 years
P14	F	Foster carer	4	M	Provisional	Since birth

1= one interview was conducted with both foster mother and father
pFAS= partial fetal alcohol syndrome, FAS= fetal alcohol syndrome, SE:AE= Static Encephalopathy: Alcohol Exposed, ARND= Alcohol Related Neurodevelopmental Disorder, Provisional= provisional diagnosis given pending further testing (i.e., awaiting further assessment, or diagnosis given as provisional due to age and the complexity in assessment amongst children younger than 6 years old), FASD= FASD diagnosis, not otherwise specified.

Table 2

Themes, exemplars and number of caregivers describing consistent experiences.

Subtheme	Exemplar quote	*n*
Major theme: Self-regulation (Behavioural)		
Hyperactivity	From the time he gets up until he has his medication and calms down, yeah- it's like a tornado hitting the house. He just races around everywhere and turns everything upside down [P9]	13
Impulsivity	Because he (8 year old) is so impulsive socially, he's probably going to be getting himself into strife. His mates know that if they ask him to do something that he will do it [P2]	13
Risk taking behaviours	I've described it as the monkey driving the bus… and I've also given the analogy to people that his thinking processes are slow like Winnie the Pooh in a Tigger body. He can bound across the road before he realises that there is a car coming, or that it's not a great idea. [P14]	8
Taking things which don't belong to them	He has particularly light fingers; in the sense that he has this… he's a great little thief. I think like an impulse thing. He has no idea like that does not belong to me, I should not touch it, or I've been told not to touch it. [P3]	11
Major theme: Self-regulation (Emotional)		
Tantrums or meltdowns	It could be anything. He could be sitting there, playing around, talking not a problem, and away he goes. It's like living with a stick of dynamite mate; you never know when it's going to go off. [P9]	11
Managing transitions	I think she had a lot of trouble transitioning from one thing to another. Like huge, I'm talking… moving from playdough to paint, she would just throw herself down, or have a great big tantrum… [P13]	8
Major theme: Self-regulation (Attention)		
Easily distracted	They have trouble being focused. You've got to prompt them and remind them. I don't know… just constantly. But they just side-track so easily, and just start doing something else. You can't keep them where they are meant to be. It's a constant struggle. [P12]	11
Needing external support	Because she can't focus, and she knows she can get away with just drifting off because somebody is not watching her all of the time. She just gets lost in her little world, and the teacher will bring her back to reality. [P5]	5

Major theme: Cognitive abilities (Learning and memory)		
Difficulty understanding consequences	She will not understand the consequence - if you throw something, it will be taken away, if you hit somebody you will be removed- stuff like that she still will not get it. We try to restrict anything that excludes her because she cannot understand if she is being excluded for any reason… but for the most part, it's generally removal of an item. But if the consequence is too long, and not immediate, we can't. She won't get it. So, it has to be an immediate consequence. [P11]	12
Working or short-term memory	You can't give her more than one instruction at once. You can't say "brush your teeth, put your pajamas on, go to the toilet", or whatever order you give it in. You have to give her one "brush your teeth" and then you have to go and make sure she's actually doing it, because if she goes out into that hallway and sees something else, or suddenly remembers something else she wants to do, she's gone and you can't… you have to chase her and say "Right, get back here, come back and do this, what were you just told to do?" [P4]	11
Forgetting previously learnt information	He's (6-year-old boy) expected to learn his sight words, we've been on the same twelve sight words since the beginning of last year. And every night we can read them and by the fifth time we've read them he can tell them to me but by the next day he has no idea. Whereas other children are on number 200, we are still on the first twelve" [P6]	5
Need for repeated exposure	Being told something and then just staring at you like they have never heard that before "you've never said that before" [P2]	8
Major theme: Cognitive abilities (Academic abilities)		
Academic achievement generally	She's (11-year old) probably anywhere between year one and three, so they are giving her work content between one and three. Very little year three stuff. [P5]	10
Maths difficulties	Maths is a real struggle, all mathematical concepts. They are not too bad when they cook with him but again you've got visual cues and you are discussing with them you know two-thirds what it's going to look like if we cut this cut into three pieces and where we went to fill up to but if that was a sum on a piece of paper, they wouldn't be able to picture that at all. [P2]	10
Literacy	The ten-year-old is really doing prep which is five or six-year-old language and literacy level [P6][1]	8
Major theme: Adaptive functioning (Communication and language skills)		

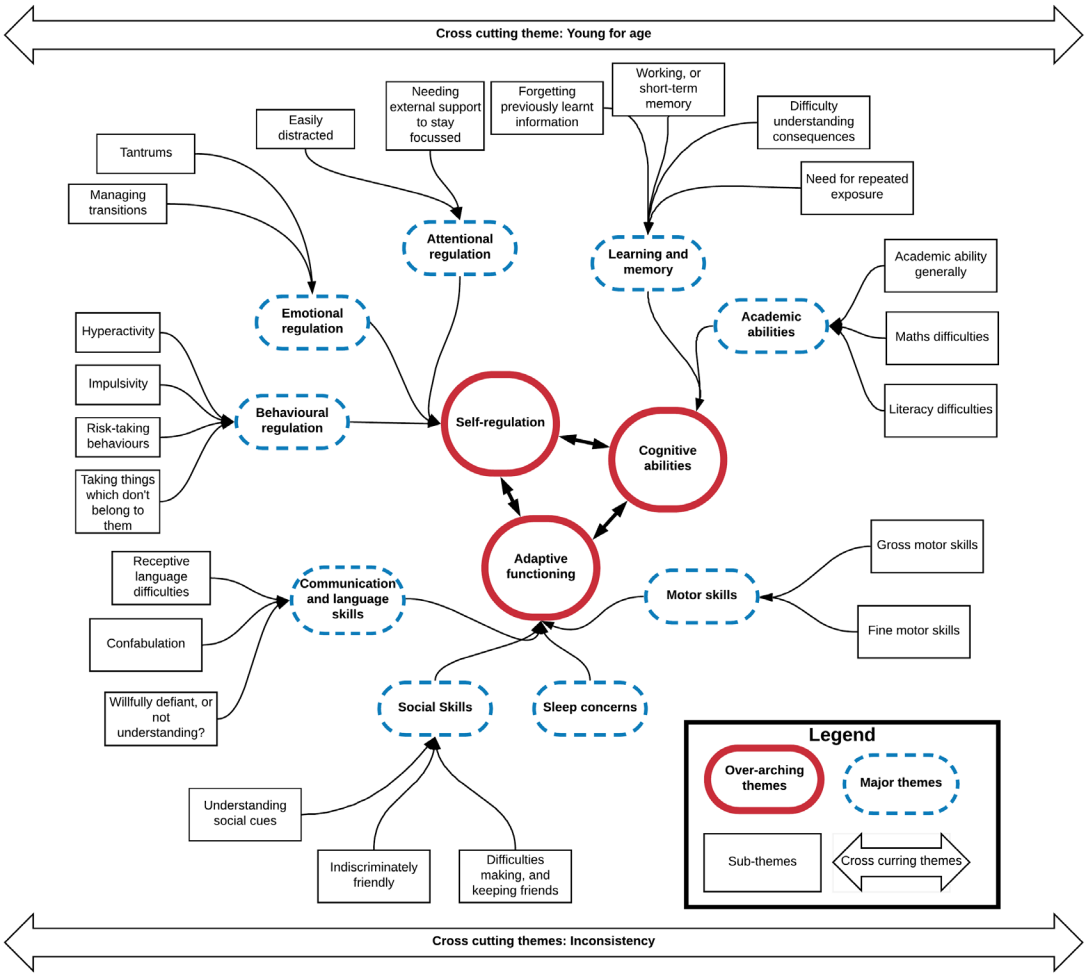
Receptive language difficulties	He can repeat back to me exactly what I've said. But to actually have comprehended that that is what's to happen, that doesn't happen frequently. [P7]	9
Willfully defiant or not understanding?	It's hard for us to determine in a situation "are you being defiant, or deliberately… or you can't understand what I'm saying", or do you… he's got a poor working memory, so do you genuinely not remember what I told you, or is it really a blank? You know like, I think he's a little bit cluey, and I think sometimes he can… I think sometimes he can manipulate the situation a bit. But it's just really hard to know what is bad wiring and what's genuine and what's pulling our leg, you know? [P3]	7
Lying and confabulation	I've had people that had had her in a car with them and she said to me that "Oh she was telling me some fantastic stories in the car", and I said "Oh, I'm sure she did". I didn't even ask, I just went I'm sure she did. Take it with a grain of salt, 'cause believe me she thinks that dreams, or things that she has thought about are actually real memories. She doesn't know one from the other. [P4]	11
Major theme: Adaptive functioning (Social skills)		
Difficulties making and keeping friends	Her friendships change constantly, and they have mentioned that at school several times, that you know she doesn't seem to be able to keep friendships and they keep having to have these friendship conversations with her about how you treat friends and that kind of thing. [P4]	11
Understanding social cues	He is very social, but it's just trying to teach him those social skills of when you are getting in someone's personal space which he doesn't understand. But he doesn't like it if they do it to him- he gets quite upset if someone gets in his personal space. [P10]	9
Indiscriminately friendly	[P13F]: Incredibly friendly. [P13M]: But once again it is inappropriate, the full on from first meeting. [P13F]: Well, she hasn't read the cues to see if she can trust this person. She's just gone all in and… It's quite scary to see, and it comes back to a vulnerability when she gets older. [P13]	7
Major theme: Adaptive functioning (Motor skills)		
Gross motor skills	Right from the word go I did a lot of stuff with him, but even with that his balance is still a problem. He couldn't ride a bike, until he was probably about grade two, grade three until he could not have training wheels. And then, swimming he still cannot come over to breathe- to the side- without rolling all the way over [P7]	8

Fine motor skills	She loves to help, but the skills just aren't there to be able to help. There is no fine motor control to be able to… say she really wants to help with the tea, well there is very limited things in what she can do because she can't actually do so many things. So what we actually end up doing is just hand over hand- "Oh you want to chop up the vegetables, alright then" so she can get the vegetables out but she wouldn't be able to do what … any other… child her age could do [P13]	12
Major theme: Adaptive functioning (Sleep concerns)		
Sleep concerns	As he started getting to around 4-years-old or something, the sleeping at night was shocking right up until he was diagnosed [at 7 years old], it would take me until two or three o'clock in the morning to get him to sleep. That was doing every single possible thing you know, to calming music, to no stimulation whatsoever before bed, to sitting with him with stories. He just could not sleep. And then, I would have to wake him up at 7[am] for school and he would just be, oh my god, so bad. [P1]	10
Cross-cutting theme: Young for age		
	His behaviour is very young- very childlike. So, he doesn't act out situations like a ten-year-old should, so he is probably more like a seven-year-old, that kind of age group. [P3]	13
Cross-cutting theme: Inconsistency in behaviour and strategies		
	It's almost like a can't really win situation sometimes. If you are silly enough to think that he has recalled it [holding hands when crossing the road] this time… there is a good chance that he hasn't. And the days where he does, he gets really upset that you are [holding his hand when crossing the road]… so getting out of the car to go into Kindergarten can be, you know, a life or death experience on a daily basis. [P14]	7

1- P6 reported her two children with FASD to be 6, and 9 turning 10 in a month at the beginning of the interview. This quote is presented verbatim.

Figures

Figure 1. Diagrammatic representation of the themes identified in thematic analysis.



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Supplementary Files

Supplementary file 1: Semi-structured interview guide

Prior to interview commencing

- Explain the purpose of research
- Re-confirm that the participant is interested and willing to participate
- Read information and consent sheet, answer any question. Indicate participant's consent.
- Start recorders after consent, and let participant know that the recorders are running

Stage 1: Introduction and demographic details

- What is your relationship to your child/ren with diagnosed with or suspected of having FASD?
- What gender is/are these child/ren
- How old are these children?
- How old were children when the diagnosis was conferred?
- What diagnosis did your child receive?

Stage 2: Body of the interview

- What are some of the cognitive and behavioural difficulties that your child experiences?
- How are their relationships with other children, peers, or siblings?
- How are they performing in school?
- When did behaviours being to emerge, and what led you suspect that this child may have FASD?
- What areas of strength do they display?
- Is there anything else you would like to add?

Stage 3: Closing off the interview

- Make arrangements for sending transcript for additional comments, and gift card

Supplementary file 2: Consolidated Criteria for Reporting Qualitative Research (CORE-Q)

Adapted from Tong, Sainsbury, and Craig (2007)

No	Item	Guide questions/description	Page reported on, or additional information
Domain 1: Research team and reflexivity			
Personal Characteristics			
1.	Interviewer/facilitator	Which author/s conducted the interview or focus group?	Pg 11
2.	Credentials	What were the researcher's credentials? *E.g. PhD, MD*	Pg 11
3.	Occupation	What was their occupation at the time of the study?	Pg 11
4.	Gender	Was the researcher male or female?	Pg. 11
5.	Experience and training	What experience or training did the researcher have?	Pg. 11
Relationship with participants			
6.	Relationship established	Was a relationship established prior to study commencement?	Pg. 9
7.	Participant knowledge of the interviewer	What did the participants know about the researcher? e.*g. personal goals, reasons for doing the research*	Pg. 9
8.	Interviewer characteristics	What characteristics were reported about the interviewer/facilitator? e.g. *Bias, assumptions, reasons and interests in the research topic*	Pg. 11
Domain 2: study design			
Theoretical framework			
9.	Methodological orientation and Theory	What methodological orientation was stated to underpin the study? *e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis*	Pg. 10-11
Participant selection			
10.	Sampling	How were participants selected? *e.g. purposive, convenience, consecutive, snowball*	Pg. 9
11.	Method of approach	How were participants approached? e.*g. face-to-face, telephone, mail, email*	Pg. 9

No	Item	Guide questions/description	Page reported on, or additional information
12.	Sample size	How many participants were in the study?	Pg. 8, table 1
13.	Non-participation	How many people refused to participate or dropped out? Reasons?	Pg. 8
Setting			
14.	Setting of data collection	Where was the data collected? e.*g. home, clinic, workplace*	Pg. 9
15.	Presence of non-participants	Was anyone else present besides the participants and researchers?	Pg. 10.
16.	Description of sample	What are the important characteristics of the sample? *e.g. demographic data, date*	Table 1
Data collection			
17.	Interview guide	Were questions, prompts, guides provided by the authors? Was it pilot tested?	Supplementary table 1
18.	Repeat interviews	Were repeat interviews carried out? If yes, how many?	No repeat interviews
19.	Audio/visual recording	Did the research use audio or visual recording to collect the data?	Pg. 10
20.	Field notes	Were field notes made during and/or after the interview or focus group?	Pg. 10
21.	Duration	What was the duration of the interviews or focus group?	Pg. 9
22.	Data saturation	Was data saturation discussed?	Pg. 10
23.	Transcripts returned	Were transcripts returned to participants for comment and/or correction?	Pg. 9-10
Domain 3: analysis and findings			
Data analysis			
24.	Number of data coders	How many data coders coded the data?	Pg. 11
25.	Description of the coding tree	Did authors provide a description of the coding tree?	Figure 1
26.	Derivation of themes	Were themes identified in advance or derived from the data?	Pg. 11
27.	Software	What software, if applicable, was used to manage the data?	Pg. 10
28.	Participant checking	Did participants provide feedback on the findings?	Participants were invited to provide feedback on their transcripts or provide additional

No	Item	Guide questions/description	Page reported on, or additional information
			comments; however, no comments were received (Pg. 9-10). Further, some participants from this study were involved in subsequent validation studies based on the themes identified.
Reporting			
29.	Quotations presented	Were participant quotations presented to illustrate the themes / findings? Was each quotation identified? e.*g. participant number*	Table 2
30.	Data and findings consistent	Was there consistency between the data presented and the findings?	Table 2
31.	Clarity of major themes	Were major themes clearly presented in the findings?	Table 2, pg.12-20
32.	Clarity of minor themes	Is there a description of diverse cases or discussion of minor themes?	Pg. 12-20

Chapter 3B Clinician Interviews

Individuals with FASD experience a range of cognitive and behavioural difficulties, as highlighted in Chapter 2 and 3A. The aim of this thesis is to develop a screening tool to identify children who display the cognitive and behavioural impairments consistent with FASD. Qualitative research with a range of stakeholders is central to the development of measures to ensure that the content of the tool covers the domains that are important to include, and as an important source of items (Haynes et al., 1995; Streiner et al., 2015). In addition to the interviews with caregivers as presented in Chapter 3A, a series of interviews were conducted with clinicians who regularly work with children with FASD in assessment, diagnostic, or therapeutic roles.

While previous research highlights a lack of FASD awareness and knowledge amongst clinicians and health practitioners generally (Domeij et al., 2018), no studies have been conducted exploring the strengths and difficulties of children with FASD from the perspectives of clinicians who regularly work with these children. Clinicians have an important perspective to offer, as they may be seeing multiple children impacted by cognitive and behavioural characteristics and are able to report commonalities and differences across children with FASD. One existing tool intended to capture the behavioural profile of children with FASD- Personal Behaviour Checklist-36 (formerly the Fetal Alcohol Behaviours Scale)- was developed from extensive clinician experience (Streissguth et al., 1998), however qualitative studies detailing this input appear to have not been published. This chapter sought to examine the cognitive and behavioural functioning of children with FASD, from the perspectives of clinicians who regularly work with children in diagnostic and assessment roles. Clinicians from a range of professional backgrounds (e.g., paediatricians, psychologists, occupational therapists, and speech

pathologists) were sought, given the importance of the multi-disciplinary team in formulating a diagnosis of FASD (Bower & Elliott, 2016).

Methods

Procedure

Ethical approval was obtained from the University of South Australia Human Research Ethics Committee (Protocol number: 0000036058. See Appendix B). Ethics approvals were limited to Australian participants, hence potential overseas participants were excluded. A purposive sampling approach was taken to identify Australian clinicians (e.g., medical doctors and allied health professionals) with expertise in the assessment, diagnosis, and therapeutic roles with children with FASD. Clinicians were eligible to participate if they were currently working in a diagnostic or therapeutic role with children with FASD.

Given the focus of the interviews on identifying the cognitive and behavioural profile of children with FASD, ensuring clinicians had experience in the diagnosis and assessment of FASD was vital. There are current extensive efforts underway to improve the training of clinicians in Australia, however, the pool of potential participants was limited to those who had received training in, and who were conducting assessments, diagnosis or therapeutic services with children with FASD. Efforts for recruitment focused on recruitment through the FASD Service Hub Australia service directory. The service directory is a web-based directory of clinics and clinicians, who can nominate to be listed on the service hub. The nomination process involves detailing their experience in FASD diagnosis or assessment, or their work in therapeutic roles with individuals with FASD.

Clinics listed on the FASD Hub Australia service directory (https://www.fasdhub.org.au/services/) were contacted and asked to distribute recruitment flyers

to their staff (see Appendix C). Sole practitioners also listed on the service directory were contacted and informed of the purpose of the study and invited to participate. At the time of recruitment, there were 30-40 clinics and clinicians listed on the service directory. Significant attempts were undertaken to recruit clinicians given the importance of the perspectives of clinicians. Clinicians and clinics were initially contacted via email to advertise the study. Up to three follow up phone calls were made to clinics to encourage clinic staff to distribute materials to clinicians. Further, a convenience sample of clinicians from the Australian FASD Centre for Research Excellence were contacted via the research team. Recruitment was undertaken between June and November 2017. Clinicians were informed of the purpose of the study and invited to participate in a semi-structured interview. No incentives were offered for clinicians to participate in the interviews.

Participants

Clinicians were eligible to participate if they were currently working in a diagnostic or therapeutic role with children with FASD. Eight clinicians contacted the research team, with five clinicians participating in interviews. Non-participation was related to the individual being unable to be contacted on follow up or having received FASD training and working in a FASD assessment role but yet to have conducted assessments in the role. Participating clinicians were asked about their professional background and experience, before discussing common referral concerns, assessments typically undertaken with children with FASD, the domains of functioning that would be useful to capture in a screening tool, the ages of children with which they typically worked with, and whether there were any gender differences in the cognitive and behavioural functioning of children with FASD that they had observed (see Appendix D for the semi-structured interview guides).

Data Management and Analysis

As per the caregivers' interviews (Chapter 3A), clinician interviews were all conducted via telephone in a private office. Interviews were audio-taped with the clinician's consent and were transcribed by the researcher. A target was set for 15 interviews with clinicians of children with FASD, however this target was not achieved, and clinicians' interviews did not achieve saturation (Brod et al., 2009). The clinician interviews were analysed using thematic analysis, following the same procedure described in the caregiver interview study (Braun & Clarke, 2006).

6. The first author transcribed all interviews, checked transcriptions for accuracy, and read and re-read each transcript.
7. Initial codes were generated. Initial codes were discussed in regular meetings between the first author and the research team.
8. Codes were collated into potential themes. Existing research was drawn upon to guide the collation of codes into themes, including the Australian FASD diagnostic guidelines (Bower & Elliott, 2016), and reviews of the cognitive and behavioural functioning of children with FASD (Kodituwakku & Kodituwakku, 2014).
9. The themes were reviewed and revised and checked against both the coded extracts and the original data set. For the purposes of inter-rater reliability, a random selection of five transcripts were reviewed against the coding by a second coder from the research team.
10. The codes were further refined, named, and defined.

Analysis was initially undertaken examining transcripts from caregiver and clinician interviews together to examine the common difficulties experienced by children with FASD. All analysis was conducted using NVivo 12 (QRS International, 2018).

Results

Clinicians interviewed included two paediatricians (one of whom identified as a developmental paediatrician), one clinical neuropsychologist, one speech pathologist, and one occupational therapist. Clinician interviews lasted on average 26 minutes (range = 17-30 minutes). Demographic details are provided in Table 3.1.

Table 3.1

Demographic data

ID	Specialism	Years experience	Setting	Ages of children
C1	Occupational Therapist	Unspecified	Private practice, and conducting assessments for multi-disciplinary clinics	4 to 18 years
C2	Developmental Paediatrician	6	Developmental clinic	Works with children across age ranges, but spoke to experiences with primary school aged children (e.g., 6 to 8 years)
C3	Clinical Neuropsychologist	10+	Diagnostic and assessment, large proportion with youth justice	4 to 12 years
C4	Speech Pathologist	3	Diagnostic and assessment, primarily youth justice	Primarily ages 10 to 17 years
C5	Developmental Paediatrician	10+	Developmental clinic	Unspecified

Cognitive and Behavioural Profile

Cognitive and behavioural functioning of children with FASD was analysed in conjunction with the caregiver data. Consistent with the themes identified in the caregiver interviews, themes are arranged into three over-arching themes: self-regulation comprised of

behavioural regulation, emotional regulation, and attention, cognitive ability comprised of academic ability and learning and memory, and adaptive behaviour comprised of social skills, communication and language skills, and motor skills. Clinicians also identified several other themes relevant to the cognitive and behavioural functioning of children with FASD including the age at which difficulties emerged, and the overlap between FASD and trauma experiences.

Self-regulation.

Three themes were identified under the over-arching theme of self-regulation; behavioural regulation, emotional regulation, and attention regulation.

Behavioural regulation.

Behavioural regulation was defined, in line with the caregiver interviews as descriptions relating to hyperactivity and impulsivity. Three clinicians reported they had observed difficulties with behavioural regulation amongst children with FASD. Clinicians' descriptions of behavioural regulation focused on hyperactivity or impulsive behaviours. Children with FASD were described as "*Not sitting still or working independently".* The speech pathologist noted that "*younger adolescents present far more problematic with behaviour ... you'll get* [Kids] *that have a real difficult time with their attention, and executive function and they are just bouncing off the walls".*

Children with FASD were reported to often have questions raised about whether their behaviours may reflect ADHD. As one paediatrician noted: "*A lot of the children for who there is a question around FASD get referred for whether they may have ADHD or there is some other learning or concentration related difficulties"*.

Impulsivity, and difficulties with inhibition, were a key concern for the clinical neuropsychologist who reported that "*definitely inhibition, I would say that's one of the markers*

every time I see students [clients in their care]*, depending on their age- that's across the pool but depending on their age".* Further, one paediatrician (C5) suggested that caregivers of children often presented with "*behavioural problems including ADHD and problems with self-regulation and behavioural problems or profiles that seem like conduct disorder or oppositional defiant disorder"*.

Both paediatricians made observations over whether gender differences in internalising and externalising behaviour may be seen in more male children being identified and referred for assessments.

> *"I think that it is less likely- and this goes to just ADHD generally- that girls present with the hyperactivity, they are more likely to present with inattention and for that reason sometimes if they are under the radar shall we say for teachers picking up and telling parents that there is a concern there".* (C5)

> *"And my sense is that they probably are less disruptive to others and less disruptive in the classroom, so that they're less likely to be used as a trigger for referral for assessment".* (C2)

Emotional regulation.

Emotional regulation was raised by two clinicians. The occupational therapist commented that when conducting assessments in their clinic, the environment around children may be over stimulating and children may be more sensitive to environmental triggers, highlighting the role of sensory processing in emotional regulation:

> *"[When] I was assessing kids in the clinics they were often heightened, or it would be quite distracting because there would be all this new equipment around them, or a bit too*

stimulating or anxiety provoking. Even just to get kids into a clinic was really tricky". (C1)

Further, it was noted that children's emotional regulation could vary substantially across days. One paediatrician (C2) reported that mental health symptoms such as anxiety and depression concerns may more frequently emerge in older children, noting that: *"the mental health stuff becomes more apparent - that they've got difficulties with poor social judgement, or perhaps a little bit more obviously unable to work independently - as they get a bit older*".

Attention regulation.

Three clinicians discussed attention difficulties among children who may have FASD. The clinical neuropsychologist and a paediatrician cited attention problems such as inattention in the classroom "*if you go back and look at their earlier trajectory they are all kids who had either been diagnosed with ADHD or have had similar sorts of difficulties in terms of their attention and concentration".* (C3)

Clinicians also discussed attention difficulties in the context of conducting assessments. The occupational therapist noted that children with FASD often need additional support when conducting assessments:

> *"So even just explaining what to do to them, you definitely have to break it down, give them a few more tries, be quite flexible and often when you are timing it, I stop counting but you let them just get a few more [tries] so the motivation continues, because they can get really exhausted".* (C1)

Similarly, attention difficulties were cited as potential contributors to memory difficulties displayed by children with FASD in the context of the assessments by the clinical neuropsychologist.

> *"Memory…Children's Memory Scale, the WRAML* [Wide Ranging Assessment of Memory and Learning]*.... Everyone uses the-…there is a big use of the WRAML Stories, and I just don't like for this population. They are too long, and they are…you kind of start loading onto the attention difficulties rather than picking up memory difficulties, so I do the primarily CMS* [Child Memory Scale] *Stories.* (C3)

Cognitive Ability

Two themes were identified under the over-arching theme of cognitive ability; learning and memory, and academic ability.

Learning and memory.

Two clinicians described children with FASD having difficulties learning from their mistakes and forgetting previously learnt information. One paediatrician (C2) noted: *"Typically it's the child who is not responding to input, so they keep making the same mistakes*," further noting that among older children: "*Some of the older kids have more difficulties with some of the more executive function-type concerns in what looks like recidivism, inability to learn from their previous exposures to peer pressure and so forth".* The speech pathologist (C4) noted that individuals with FASD appeared as though they were "*not learning from mistakes… and not learning from previous experiences".*

Academic ability.

Impaired academic abilities were discussed by most clinicians, but this was frequently in the context of behavioural difficulties experienced by children living with FASD. The paediatricians both reported children often experience academic difficulties, but children more often come to the attention of clinicians or are referred if there are concerns regarding behaviour regulation:

"Academic performance is often a reason for referral and [is] almost always associated with behavioural problems… I think a lot of kids with academic problems who don't have behavioural problems get assessed perhaps by school psychologists and or already have some kind of assessment whereas it's a kid who almost [flies] through under the radar in terms of not bottoming out in class but then their behaviour problems become such an issue that that's why they get referred".(C5)

"Otherwise there might be identified learning difficulties that have lead to some behavioural concerns, usually in the classroom".(C2)

The speech pathologist described a history of difficulties in the classroom often prompting an investigation of language difficulties: "*the big one that I look for is poor school engagement, or poor academic success and straight away think probably looking at a language disorder"*. The speech pathologist further discussed the potential for language difficulties experienced by children with FASD to impact upon children's performance in the classroom, highlighting the inter-relation between domains:

"You know- equation, multiply, subtract, all of these … words I would say can be difficult for the kids with a language difficulty because they haven't really developed that and learnt the word yet. And when you mix that with sentence structures- really complex sentence structures- umm, you know like "Jenny walked home from school. It took her 20 minutes, but she went via somewhere else la la la la la" The kid has to one- understand the actual sentence, and then also lay on top of that the maths vocabulary, and that can often be challenging for them". (C

Adaptive Functioning

Three themes were identified under the over-arching theme of adaptive functioning; motor skills, communication and language skills, and social skills.

Motor skills.

The occupational therapist spoke extensively about the difficulties in gross and fine motor skills experienced by children with FASD, highlighting a range of motor skills difficulties;

> *"Definitely lots with their motor co-ordination, quite a lot of balance issues. Fine motor skills is a really big one, so manual dexterity, visual-motor integration, fine motor precision usually are quite heavily impacted as well. Gross motor… strength usually is quite a big one, like really poor postural strength (C1)*

Further, difficulties with motor planning and coordination (e.g., the coordination the movements of the body and planning out an action, such as planning the sequence of putting on a pair of pants) was highlighted as a potential area of weakness: "*the motor coordination bit I definitely find that they score quite significantly lower in that one* [than anyone else]. *And you can kind of predict it before they even start it that they aren't going to go very well in i*t."

Two clinicians (occupational therapist and clinical neuropsychologist), highlighted that visual perception skills were a relative strength among several children they see:

> *"they are usually sometimes stronger in their visual perception skills and we know that that population have much stronger visual-spatial skills anyway, so are they performing at what their level would have been, or is it just a relative strength amongst other things, even though it may have been impacted"* (C3)

While gross motor skills- in particular balance, and postural strength- were noted as a common difficulty among children with FASD, participation in extra-curricular activities was described as an important protective factor in the development of motor skills:

> *"It depends as well, some might be really inactive and not participate in any other extra-curricular activities, so there is that kind of issue as well. I've got some kids that don't do anything outside the school, so they don't have any chances to develop their skills, as well as the impact on the brain".*

The speech pathologist briefly noted that reduced motor speech skills were also present in a number of older children and adolescents:

> *"We also see motor speech and articulation difficulties, so by the time I see them, which is when they are adolescence… they haven't developed their speech skills but some of them still have reduced motor speech skills"*

Communication and language skills.

The speech pathologist spoke extensively about the language difficulties experienced by children with FASD, noting work that has primarily been conducted with older children (10 years old, or older). The speech pathologist noted several language difficulties among children with FASD including:

> *"Receptive language disorders, poor vocabulary and understanding of words, having difficulties following instructions, not understanding the gist of information that they might be given, like a long list of information that they might be asked to follow but only picking out bits and pieces.*

Although typically working with children at the older end of the range, early language skills including phonological processing were reported to occasionally be a difficulty for children

with FASD. Language skills were also noted to impact upon other domains, including social skills, with language difficulties reported to contribute to them struggling to maintain friendships, and language difficulties contributing to impairments in academic skills including maths, due to the abstract nature of language around maths. Language skills were also briefly raised by the clinical neuropsychologist in relation to the presentation of children with FASD: "*That* [inhibition] *seems to be one of the markers across the board with the kids that I see. And language as well".* It was noted that language skill impairments may be impacted by experiences of neglect:

> *"And so if you get neglect of environmental stimuli, then you are going to be getting neglect of language interaction, which may impact on language skills which are independent of alcohol exposure in utero which can be very hard to determine".*

Social skills.

It was noted by one paediatrician that one of the concerns relating to the identification of children with FASD was emerging social skills difficulties in middle childhood;

> *"It's more obvious as we head towards that middle childhood, where it is more the social interactions are more difficult, where we are overly friendly, we haven't worked out what is right and what's not right, where we will sort of go along with whoever will get us into trouble or whoever will lead us astray for example".*

Further, the speech pathologist noted that language difficulties- and the impact on social skills and social communication- may contribute to children with FASD struggling to maintain friendships: "*Social skills difficulties … social skills and social communication. So we'll see… kids that… struggle maintaining friends, or they are easily led by people".*

Additional Themes

Clinicians raised a number of themes relevant to the identification of children and young people who have FASD, or conducting assessment for FASD. Clinicians discussed when difficulties became apparent, and the role of post-natal experiences in influencing the cognitive and behavioural presentation of children with FASD.

When difficulties become apparent.

With regard to when difficulties become apparent; one paediatrician (C2) reported there was a less clear picture of early developmental milestones, but as children were heading into school, difficulties became more noticeable:

> *"And it's sort of middle childhood period, where the conceptualisation of what is right and what's wrong, whether you should lie, what happens there in terms of… so I think that's why you aren't seeing those really early developmental milestones, so for example, lying isn't really something that you'd see before the age of four. So not really the more traditional gross motor skills, fine motor skills levels, it's more sort of as you are heading into school and the interaction and the, I guess, the interactions with others that it's more noticeable"* (C2)

Further, clinicians noted that children referred for FASD assessment often had learning or behavioural difficulties in the classroom.

Contribution of experiences such as trauma.

Clinicians discussed the role of trauma in the presentation of children with FASD. The clinical neuropsychologist stressed the importance of understanding both pre-natal and post-natal exposures of children to effectively manage their behaviour:

"*Many of these kids present with negative psychosocial histories, so it's hard to tease out some of these issues that are specific to in utero exposure to alcohol rather than their psychosocial circumstances."* (C2)

The clinical neuropsychologist highlighted the importance of ensuring there is a comprehensive assessment of children's psychological health in making diagnosis and treatment plans:

> *"I think for these kids there is an acknowledgement of trauma, anxiety, a whole range of conduct presentations, however for the kids that I've seen, they haven't been systematically assessed, so that is I think a missing puzzle* [piece] *with these kids. There needs to be a very clear understanding of their psychological health, or not, to contribute to this diagnostic picture.* (C3)

Further, one paediatrician noted that with children presenting with ADHD-type behaviours and impulsivity, further assessment was often required to tease out the role of both PAE and childhood trauma. Lastly, the occupational therapist commented on the role of trauma in sensory processing noting that: "*often I find that it* [sensory processing difficulties] *does come up quite high. If there is that side of trauma as well, I will put that in to… I guess it shows up with trauma and the FASD more so.*

Discussion

The qualitative studies presented in Chapters 3A and 3B sought to identify the cognitive and behavioural difficulties experienced by children with FASD, from the perspective of caregivers and clinicians working in assessment, diagnostic, and therapeutic roles. Fifteen caregivers of children with FASD and five clinicians from a range of professional backgrounds participated in interviews exploring the difficulties experienced by children with FASD, common

referral concerns, and differences across ages and genders. While the numbers of clinicians were small, they described cognitive and behavioural difficulties that were largely consistent with that of the caregivers and provided additional clinical insights regarding their work with children with FASD and similar conditions.

The interviews with caregivers identified a range of common cognitive and behavioural challenges experienced by children with FASD, including challenges broadly grouped into self-regulation (i.e., behavioural regulation, attention regulation, and emotional regulation), cognitive abilities (i.e., academic and learning and memory), and adaptive functioning (i.e., communication and language, social skills, young for age). Clinicians identified as similar pattern of difficulties. Clinicians commonly identified difficulties with behavioural regulation, cognitive ability (particularly difficulties learning from mistakes). Reflecting their specialisms, the occupation therapist and speech pathologist spoke extensively about difficulties in motor skills and language, respectively. While the same is true of the clinical neuropsychologist, the domains of impairment associated with FASD that are typically assessed by psychologists are far broader (e.g., attention, learning and memory, executive functioning, cognition and cognitive ability). It is important to note that the clinicians were mindful to limit their responses to the areas that they were most knowledgeable in.

The clinicians also raised several issues that had been touched on during the caregiver interviews. Clinicians described the impact of trauma on the behavioural presentation of children with FASD, noting trauma was often a contributing factor to the cognitive and behavioural profile of children living with FASD. Further, several clinicians noted they worked extensively in child protection and youth justice populations, where there may be experiences of trauma and

early life adversity. Areas of comorbidity and confounding are explored in greater detail in the subsequent chapters.

Transition periods such as starting school were highlighted as an important time for identify children who may be experiencing difficulties. With regards to the age at which children who were experiencing difficulties were identified, one clinician noted the interactions with peers as especially important in identifying children who may have FASD. It was reported that social development in middle childhood was an important sign. Clinicians noted that children referred for assessment often had learning or behavioural difficulties in the classroom. These findings suggest that routine screening points – such as on-entry school assessments – may represent an important opportunity to screen and refer children at risk of developmental disorders, including FASD. Further, the clinicians noted that teacher reports of classroom behaviour (e.g., on-task performance) were often useful in the assessment and diagnosis of children. Notably, one paediatrician reported that referrals for assessments from school reflected behavioural regulation difficulties impacting upon classroom performance.

Differences Between Caregivers and Clinicians

The cognitive and behavioural functioning difficulties described amongst children with FASD were broadly consistent between caregivers and clinicians. However, caregivers and clinicians appeared to describe some differences in relation to emotional regulation. While caregivers described a range of behaviours relating to the emotional regulation of children with FASD, these were different to the emotional regulation themes noted by the clinicians. Caregivers spoke to themes which may be more reflective of situational or context specific such as tantrums and meltdowns, and difficulties with children managing transitions between activities. One clinician described that children with FASD may become overwhelmed in the

testing sessions conducted at the clinic. Clinicians also discussed the emergence of mental health concerns, considered emotional regulation under the Australia diagnostic guidelines definitions of emotional regulation, which were not raised by caregivers in relation to FASD. Comorbid exposure to trauma may place children with FASD at greater risk of mental health concerns (Ritter, 2019). Clinicians, however, may be more informed of mental health issues.

Caregivers and clinicians may have a different frame of reference for behavioural concerns. Caregivers have more opportunities to witness behaviours across settings and understand the situational specificity of behaviour and whether there are particular triggers for behaviour. Other themes which were discussed by caregivers and not clinicians included difficulties with sleep, consistent with previous research which has highlighted that sleep concerns among children with FASD may be missed by health care providers (Ipsiroglu, McKellin, Carey, & Loock, 2013).

Clinicians' Areas of Specialisation

The clinicians interviewed represent the range of professions that are recommended to be involved in the assessment and diagnosis of children with FASD in Australia (Bower & Elliott, 2016). While the sample of interviewed clinicians could speak to the breadth of the difficulties experienced by children with FASD, it is important to note that the clinicians often limited their comments to their areas of expertise. For example, the occupational therapist and speech pathologist spoke extensively to the difficulties that children with FASD experienced in relation to motor skills and speech and language, respectively. Although comments were limited to the domain focussed upon by their professions, clinicians were also able to speak to specific impairments in greater depth and offer inferences of some of the processes underlying the difficulties. While interviews were conducted with at least one of the professional groups

recommended to be involved when a diagnosis of FASD is being formulated (i.e., paediatrician, psychologist, occupational therapist, and speech pathologist), having such small numbers is a limitation in terms of ensuring data saturation was reached. Lastly, several clinicians noted that a sizeable proportion of the referrals to them came through either child protection or youth justice as noted in Table 3.1, and while the clinicians spoke generally about children with FASD, this may be a source of bias in the interviews. Individuals with FASD presenting to youth justice may represent children whose cognitive and behavioural functioning is more impaired and coupled with trauma. Experiences of both FASD and trauma appear to be associated with poorer outcomes (Price, Cook, Norgate, & Mukherjee, 2017). Further, experiences of trauma puts children with FASD at greater risk of contact with youth justice (Fast & Conry, 2009). While individuals with FASD may make up a significant proportion of their caseload- recent research has found that 36% of children in youth detention in Western Australia had FASD (Bower et al., 2018)- it may be a skewed representation of all children with FASD. This possibly skewed FASD representation is tempered by the inclusion of the literature review (Chapter 2) and caregiver interviews (Chapter 3A) for broader perspectives.

Limitations

While the inclusion of the perspectives of clinicians was a unique element of this study, the numbers of participating clinicians was small, thereby limiting the range of perspectives represented, as well as the opportunity to identify areas of consistency. As FASD diagnosis and assessment is in its early years in Australia, at the time of recruitment there were few clinics and clinicians with training in FASD assessment and intervention who were routinely working with children with FASD. The available data on the number of clinics and clinicians routinely diagnosing children with FASD is itself limited as it relies upon reports to be filed with the

Australian Paediatric Surveillance Unit (APSU) following a diagnosis, and there may be clinicians or clinics diagnosing FASD who are not reporting to the APSU (Zimmet & APSU FASD Investigators, 2019). Clinicians in this study noted that they have likely been working with children with FASD for years, but the condition may have gone undiagnosed due to a lack of knowledge or diagnostic clarity. It is only more recently that there has been a push towards training clinicians for FASD diagnostic and therapeutic services in Australia (Reid, White, et al., 2019). As the pool of trained and experienced clinicians widens, there will likely be additional opportunities for qualitative and other research involving clinicians.

The field of FASD has undergone a rapid expansion in Australia in recent years. At the time that the recruitment of clinicians occurred (during mid-late 2017), there was a dearth of clinicians with training and experience in the diagnosis and assessment of FASD in Australia. The list of clinicians maintained by the FASD Hub contained, at the time, approximately 30 to 40 clinics and clinicians. Substantial efforts were made to recruit as many clinicians as possible from this pool; clinics and clinicians were followed up with multiple phone calls and emails to inform them of the study and encourage participation. While several clinicians contacted the research team, difficulties in clinicians' availability prevented participation. Further, some clinicians who had received training but were yet to undertake any assessments of children with FASD were consequently excluded from participation. While the interviewees represented a range of professions, due to the small numbers of participants, the clinician interviews did not achieve saturation. Further, due to concerns of the UniSA HREC over the sensitive nature of the project, recruitment was limited to Australian caregivers and clinicians. Future research with caregivers and clinicians internationally may help to overcome the difficulties encountered with small numbers of clinicians. There are currently extensive nation-wide efforts to train clinicians

and establish greater diagnostic capacity throughout Australia, as well as the establishment of clinical networks and special interest groups within health departments across Australia.

The small clinician sample in chapter 3b potentially impacted upon the content of the pool of items that was developed in subsequent chapters. The small numbers of clinicians participating in the qualitative study and the subsequent content validation has implications for the findings, and subsequent studies should seek to conduct further interviews with clinicians to identify additional items to include in a revised version of the screening tool.

Future Research Directions

Each clinician brought a different perspective to the research, and future research should aim to ensure adequate representation of each clinical group that is actively involved in the diagnosis of FASD (e.g., paediatricians, psychologists, occupational therapists, speech pathologists). As the numbers of trained clinicians in Australia increases, there will likely be increased opportunities to recruit larger populations of clinicians for inclusion into research projects and provide opportunities to further review and refine the content of the tool. Future research could consider the use of focus groups to minimise the time demands on teams of busy clinicians, and to bring the expertise of each member of a diagnosis team to the fore. Future research could take a greater focus on the emerging difficulties experienced by children with FASD. The behaviours or concerns, and their developmental emergence, can inform screening and early intervention efforts during this period of rapid development. Lastly, future research will need to make the purpose of screening tools clear; whether tools are intended to screen universally (e.g., distinguish between children who may have FASD and typically developing peers), or in a more targeted approached (e.g., identify children how may have FASD from other conditions/comorbidities).

Conclusion

Clinicians identified several cognitive and behavioural difficulties among children with FASD. While, clinicians and caregivers spoke to broadly similar difficulties among children with FASD, there were differences noted, which may reflect the situationally specific nature of some impairments. Further, as several clinicians in this sample worked extensively with children in a youth justice population, the children may over represent those who experience greater impairments associated with FASD, or who have experienced greater post-natal adversity and trauma. The cognitive and behavioural functioning of children with FASD, as described by both caregivers and clinicians will be used to inform the development of a screening tool to identify children who display the cognitive and behavioural impairments consistent with FASD.

Summary

This chapter aimed to examine the cognitive and behavioural functioning of children with FASD from the perspectives of caregivers of children with FASD, and clinicians who work in assessment, diagnostic, and therapeutic roles. Semi-structured interviews were undertaken with caregivers and clinicians, focused on the behavioural presentation of children with FASD. Caregivers and clinicians described a range of cognitive and behavioural difficulties, with over-arching themes of Self-regulation (behavioural regulation, emotional regulation, and attention regulation), Cognitive Ability (learning and memory, and academic ability), and Adaptive Functioning (social skills, communication, motor skills, sleep concerns). The interviews informed the development of a conceptual model and item pool for the development of a screening tool to identify children who display the cognitive and behavioural impairments consistent with FASD.

Chapter 4 Content Validation

This chapter presents the item pool development and subsequent refining of the items to be included in the initial reliability and validity testing of a new FASD screening tool. Items were generated based on the conceptual framework representing cognitive and behavioural difficulties experienced by children with FASD. Cognitive and behavioural difficulties were identified through a scoping review of the literature and interviews with caregivers and clinicians (Chapters 2 and 3 as shown in Figure 4.1). The item pool was reviewed by an expert panel made up of caregivers of children with FASD and clinicians with experience in diagnostic, assessment, and therapeutic roles. Ultimately, the screening tool is intended to be completed by parents and caregivers to identify children between the ages of four and 12 who display impairments consistent with those described among children with FASD. This chapter had three aims. Firstly, this chapter reports the development an item pool containing those constructs salient to the cognitive and behavioural presentation of FASD. Secondly, this chapter describes the outcomes from the process where the item pool was reviewed by a panel of experts to ensure that the retained items showed an acceptable level of face validity (i.e., whether on the face of it, the instrument appears to be assessing the desired qualities) and content validity (i.e., whether the instrument samples all of the relevant or important content or domains), (Streiner et al., 2015). Thirdly, an initial test-ready version of the refined screening tool items which incorporated the feedback from the expert panel was prepared.

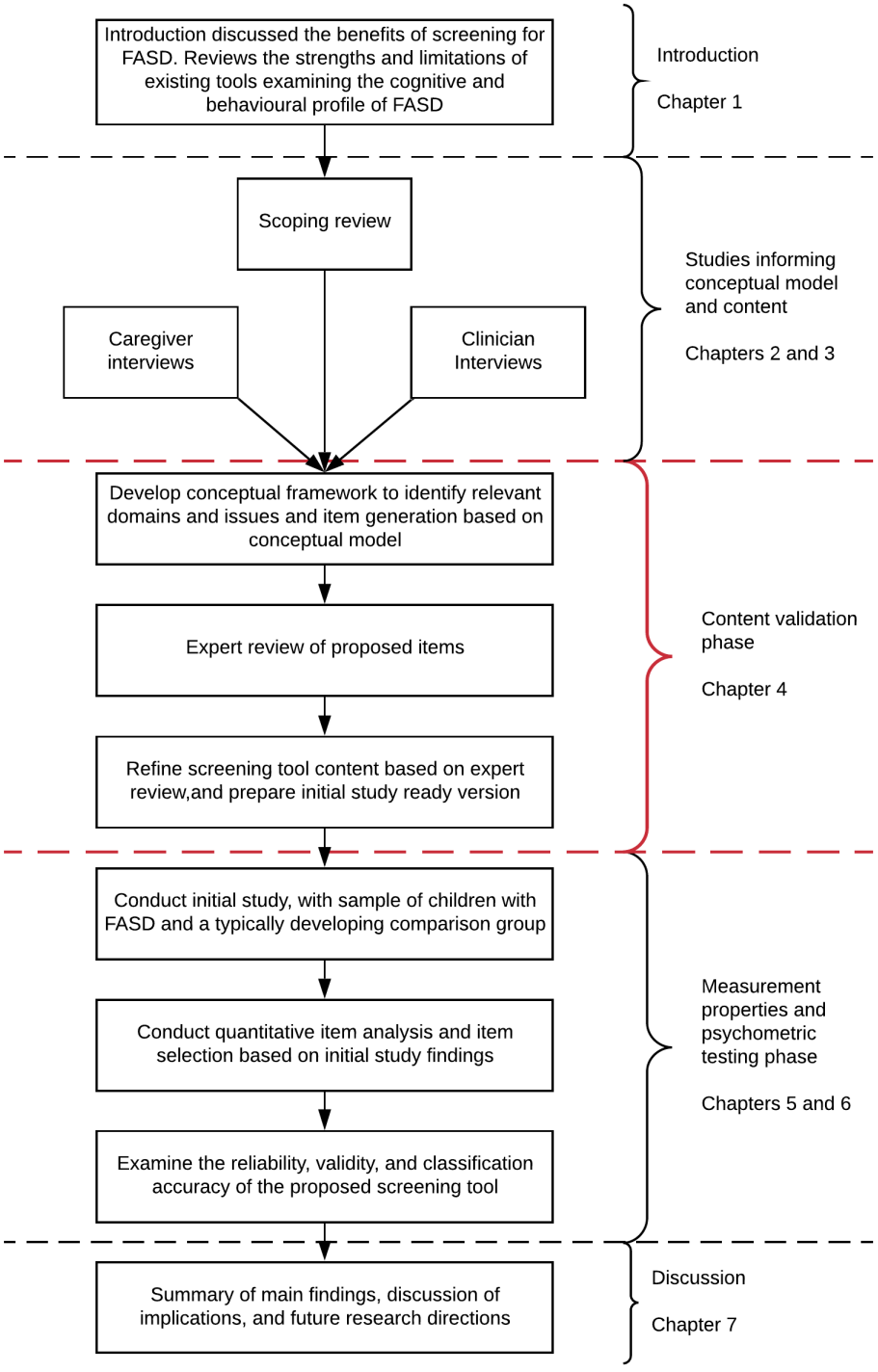


Figure 4.1. *Overview of thesis methodology and thesis structure, highlighting the present chapter. Adapted from Brod et al. (2009).*

Conceptual Framework

The findings from the scoping review (Chapter 2) and interviews with caregivers and clinicians (Chapter 3) were arranged into a conceptual framework, integrating the findings across the three sources of information (Patrick et al., 2011) The literature review and interviews both identified impairments in academic achievement, adaptive behaviour, attention, and elements of executive functioning, sleep, and sensory processing. Table 4.1 provides an overview of the functional domains and definitions generated during the coding of the interviews that informed the generation of initial items. Domains were closely aligned with the themes identified in the interviews as the focus of screening tool development was on observable and salient behaviours. In this regard, the sub-theme of "wilfully defiant or not understanding" which reflected the caregivers' interpretation of behaviour was excluded from the list of themes considered for the tool as it implied a degree of intentionality with regards to the child's behaviour but the intentionality could not be known.

Table 4.1

Domains and definitions

Domain	Definition
Self-regulation	The capacity to control and direct attention, thoughts, emotions and actions. Self-regulation is a multi-dimensional construct comprised of a number of inter-related domains, including attention, inhibitory control, working memory (McClelland & Cameron, 2012).
Behavioural regulation	The ability to control one's own behaviour, including modulating activity or arousal levels, and inhibiting inappropriate behaviours (McClelland & Cameron, 2012).
Hyperactivity	The child presents in a state of heightened activity, movement, and behaviour, that may be inappropriate for the setting.
Impulsivity	The child displays a range of actions which are poorly conceived, prematurely expressed, unduly risky, or inappropriate to the situation, and that result in undesirable consequences.
Risk-taking behaviours	The child engages in behaviour with the potential to cause injury to self or other. These are a sub-set of risk-taking behaviours.
Taking things that do not belong to them	The child is reported to take things that do not belong to them, from either inside or outside of the home.
Emotional regulation	The initiating, inhibiting or modulating of the occurrence, intensity or duration of internal feeling states, and the behaviours associated with the emotion (Eisenberg & Spinrad, 2004).
Tantrums/meltdowns	The child is described by caregivers as having meltdowns, throwing tantrums, difficulties regulating emotion, angry, explosive.
Managing transitions	The child appears to have difficulties regulating emotions when routines change either unexpectedly or with notice, or reports of transitions between activities causing heightened behaviour.
Attention Regulation	The ability to voluntarily focus and sustain attention on a task while ignoring other distractions, and shift attention as required (McClelland & Cameron, 2012).
Easily distracted	The child appears easily distracted from the current task by external stimuli (e.g., distracted by other children sitting at table).
Needing external support	The child is reported to require support from either caregiver or teachers to stay focused on a task.
Cognitive ability	The capacity involved in performing the tasks associated with perception, learning, memory, understanding, awareness, reasoning, judgment, intuition, and language (American Psychological Association, n.d.-b)
Memory	Memory is the result of a series of processes by which one forms (i.e., encodes), stores, and retrieves information. Memory includes both short-term, or working memory, and long-term memory (Schneider, 2011).
Forgetting previously learnt information	The child presents as forgetting of rules or information (e.g., spelling a target list of words) that they had previously demonstrated proficiency in.
Working, or short-term, memory	The child appears to have difficulties retaining task instructions and completing multi-step tasks (e.g., walks into room and forgets what they went in for).
Difficulty understanding consequences	The child experiences difficulties with cause and effect reasoning, repeats mistakes, does not understand consequences of actions, difficulties with punishment (e.g., forgets reason for being in a time-out).
Need for repeated exposure	The child demonstrates a need for continuous repetition of daily routines, and daily living skills for them to "stick", or for improvements to be made in working with motor skills (e.g., use of cutlery, swimming etc). Need for repeated exposure to learn new information.
Academic ability	The capacity to perform a scholastic mental act, such as the ability to learn quickly in areas of scholarship or disciplined study (American Psychological Association, n.d.-a). Reports of behaviour such as hyperactivity or impulsivity impacting on the classroom was considered under the behavioural regulation heading.
Academic ability generally	The child displays difficulties in academic performance without reference to specific content areas.
Maths difficulties	The child demonstrates difficulties understanding and applying mathematics concepts.

Domain	Definition
Literacy difficulties	The child appears to have difficulties with reading, writing, spelling, and the comprehension of written language.
Adaptive functioning	Adaptive functioning is comprised of a loosely defined set of skills associated with personal independence and social competence. Adaptive behaviour is multidimensional and covers several broad categories including: communication and language skills, social skills, and motor skills (Schalock, 2004). Sleep concerns were considered as part of adaptive behaviour.
Communication and language skills	The capacity to achieve effective communication. In addition to general language proficiency (adequate vocabulary and knowledge of syntax), effective communication involves the ability to listen and read with comprehension, to present one's thoughts clearly both in speech and in writing, to accept that the perspectives of others may differ from one's own, and to anticipate the effect of what one says or writes on listeners or readers (American Psychological Association, n.d.-c)
Receptive language difficulties	The child is reported to experience difficulties in the comprehension of what has been said to them.
Confabulation, or lie-telling behaviour	The child appears to fabricate or distort memories or demonstrates a tendency to tell lies.
Auditory and visual processing	The child appears to have difficulties or delays in the processing, or the recognition and interpretation of information taken in either auditorily (e.g., verbal instructions), or visually (e.g., written information).
Social skills	The capacity to interact socially and get along with other people including showing affection, having friends, showing and recognising emotions and assisting others. Social skills also includes interpersonal skills, avoiding victimisation, and inhibition of inappropriate or negative social behaviours (Kully-Martens, Denys, Treit, Tamana, & Rasmussen, 2012)
Understanding social cues	The child appears to have difficulties in reading social cues, and seeing the impact of their behaviour on siblings or peers,
Indiscriminately friendly	The child behaves in an overly friendly or familiar manner with strangers.
Difficulties making and keeping friends	The child demonstrates difficulties in establishing relationships with peers and/ or in maintaining friendships.
Motor skills	The capacity with gross motor (e.g., control and coordination of the large musculature used in maintaining posture and balance, and encompasses aspects of fitness and coordination involved in play, sports, and other physical activity) or fine motor skills (e.g., the control and coordination of the hands and fingers, motor skills involving control and coordination of the arms and hands, especially for object manipulation) or the acquisition of motor skills.
Gross motor skills	The child demonstrates difficulties in gross motor skills, such as balance, postures, and large body movements in playing sports.
Fine motor skills	The child displays difficulties in fine motor skills, such as difficulties with handwriting, hand-eye coordination, use of cutlery.
Sleep concerns	The child has difficulties or challenges with sleep-related behaviours including: bed-time resistance or avoidance, waking throughout the night, nightmares, and the total length of time asleep.
Young for age	Children's capacity is described as more similar to that of their younger peers. For example, described as emotionally younger than peers, behaviour in social settings more like a chronologically younger child. Also related to both a preference for, and better skills in interacting with younger peers.

Item Content Validation

To ensure that the items proposed for inclusion in the screening tool were relevant to the behavioural presentation of children with FASD, the content of the tool was reviewed by experts (Haynes et al., 1995; Streiner et al., 2015). The Content Validity Index (CVI) methodology involves having experts (e.g., clinicians, researchers, those with experience with the construct under assessment) rate the relevance of the proposed items for the concept/s that are being assessed through the tool. Indices based on the level of agreement for an individual item's relevance to the construct are calculated and used to guide the decision making on items to remove or retain from the draft version of a scale (Polit & Beck, 2006; Polit, Beck, & Owen, 2007). The subsequent sections outline the methodology for developing the initial item pool, the methodology of the expert review, and the process of refining the items.

Methods

Initial Item Pool Development

An initial pool of 300 items was developed to reflect the domains shown in Table 4.1, from the scoping review (Chapter 2), interviews with caregivers and clinicians (Chapter 3), clinical descriptions of children with FASD, and existing scales measuring the domains of interest. Caregiver and clinician interview transcripts were reviewed to identify descriptions of behaviours reflecting the domains of interest in the words of caregivers and clinicians (DeVellis, 2016). Caregivers' descriptions were often couched in the specific language of the example they were using, and edits were made to ensure that items were generalised where possible. This was supplemented by examining existing conceptual models and clinical descriptions of FASD and examining pre-existing scales. Clinical descriptions of children with FASD (e.g., Hagan et al.

(2016)) were reviewed to further source items that may not have been identified in the interviews. Lastly, existing scales of the concepts in the conceptual framework were identified as potential sources of items (Boateng et al., 2018; Streiner et al., 2015). Existing scales measuring the domains of interest were identified from the studies included in the scoping review (as they have shown validity among children with FASD, and applicability to the age range of interest), and from the pre-existing screening scales aimed to identify the behavioural phenotype of FASD as discussed in Chapter 1 (e.g., the PBCL-36 (Streissguth et al., 1998), the NST (Nash et al., 2006)), or other FASD specific measures identified in the literature review (i.e., the FAS/FAE Score (Godel et al., 2000)). To identify additional potential items, targeted searches of peer-reviewed literature were undertaken to identify proxy-reported scales measuring concepts that had not been otherwise covered (e.g., proxy-reports of memory difficulties). Where items were drawn from existing scales, individual items rather than whole scales or subscales were extracted.

At this stage of development, an item pool significantly larger than the intended final screening tool was created (DeVellis, 2016; Streiner et al., 2015). It is difficult to reinstate concepts or domains removed at the early stage of screening tool development at a later date, as such it is better to be inclusive of items during the early stages of development (Streiner et al., 2015). Therefore, in generating the item pool, the opportunity was taken to include behaviours that were identified as salient by caregivers, but which were not considered strong themes in the qualitative analysis (e.g., auditory and visual processing which was identified by four caregivers and clinicians in interviews).

The item pool was reviewed by the research team (doctoral candidate, and senior academics with backgrounds in child development, FASD and scale construction), discarding

those that were duplicates, ambiguous, double-barrelled, included jargon or value-laden words, or were overly long (Streiner et al., 2015). A total of 95 items were retained at this stage (See Table 4.3). Of the 95 items, 38 items measured behaviour related to the adaptive functioning domain, 20 items measured cognitive ability, and 37 items measured self-regulation. The 95 items were reviewed by a co-supervisor for clarity and conceptual coherence and no further changes made to the item pool. The 95 items were retained for review by the expert panel.

Item Content Validation

Expert panel recruitment.

A panel of experts consisting of caregivers of children aged between four and 12 years old with FASD and clinicians working in assessment and diagnostic roles who had previously participated in the qualitative interview study (see Chapter 3), were re-contacted to participate in the content validation study to review items proposed for inclusion in a new FASD screening tool. Polit et al. (2007) recommend recruiting between eight to 12 experts to participant in the panel. Caregivers and clinicians had provided their consent to be re-contacted for this follow up content validation study. Participants were eligible if they reported being the carer of a child with a diagnosis on the FASD spectrum or having clinical experience in the diagnosis or treatment of children with FASD. Participants were contacted via email and invited to complete the online rating questionnaire, hosted by Survey Monkey (www.surveymonkey.com). The information sheets and consent forms were presented as the first page of the online questionnaire, and informed consent was indicated by a checkbox before participants could continue with the questionnaire (See Appendix E. Ethics approval was received from the University of South Australia Human Research Ethics Committee (Protocol number: 0000036058. See Appendix B). Eight experts participated in the item review. These were six caregivers (three kinship carers, two foster

parents, and one permanent carer) and two clinicians (one paediatrician and one speech pathologist) (see Table 4.2 for demographic details). Further, while all clinicians participated in the previous qualitative phases, responses were only received from two of the five clinicians.

Table 4.2

Demographic table

Participant ID	Relationship to child	Years' experience with FASD	Number of children with FASD in their care	Child/ren diagnosis	Age of child/ren with FASD	Proportion of items endorsed
1	Permanent Carer	7	2	Fetal Alcohol Syndrome, Alcohol Related Neurodevelopmental Disorder	9, 12	76.8
2	Foster parent	11	2	SE:AE	7, 11	86.3
3	Kinship carer	6	1	FASD, not otherwise specified	6	78.9
4	Foster parent	4.5	1	FASD, not otherwise specified	6	91.6
5	Kinship carer	10	1	FASD, not otherwise specified	10	91.6
6	Carer [a]	Unknown	1	Unknown	Unknown	63.2
7	Clinician (paediatrician)	7	-	-	-	25.3
8	Clinician (speech pathologist)	4	-	-	-	85.3

Note. SE:AE = Static Encephalopathy: Alcohol Exposure.
[a] One caregiver (Participant 6) did not complete demographic questions.

Content validation index procedure.

Following guidelines for the CVI (Polit & Beck, 2006; Polit et al., 2007), a questionnaire was created containing a brief demographic section and the 95-items proposed for inclusion in the screening tool. All members of the expert panel were presented with an early version of the mind map showing the domains identified as salient to the behavioural presentation of children with FASD identified in the interviews with caregivers and clinicians (See Appendix E). Experts were asked to provide feedback if they felt there was anything missing from the model. Feedback

was only received from one participant (Speech pathologist) on this question who provided additional information on the themes relating to communication. Based on this feedback additional items were considered for inclusion in the screening tool (see section 3.2.2 for full details) Seven of eight experts had no comments on content or refinements to incorporate into the model.

Expert panel members were asked, “The research team have developed a list of items which we consider relevant for inclusion in a screening tool to assist caregivers, teachers, and clinicians identify children who may be at-risk of having FASD from the ages of four to 12. We would like your feedback on whether or not you consider these items to be relevant”. Participants were then instructed to read each of the 95 items carefully and rate each item on a four-point scale where 1 = “not relevant”, 2 = “somewhat relevant”, 3 = “quite relevant, but needs rewording”, and 4 = “highly relevant”. Experts were also provided a free-text comment box for each item and asked to use the comment box should they wish to make a comment on the item. Qualitative responses were considered by the research team during the process of revising the retained items.

Data analysis.

The CVI (Polit & Beck, 2006) was used to quantify expert judgements on the proposed items. The CVI measures the level of agreement between experts that an item is relevant to the phenomenon being measured. Similarly, the modified kappa (k^*) measures the agreement among expert judges that an item is relevant, adjusting for chance agreement. Following the conventions for calculating the CVI, responses of 1 = “Not relevant” or 2 = “Somewhat relevant” were collapsed into a *Not Endorsed* category, and responses of 3 = “Quite relevant but needs

rewording" and 4 = "Highly relevant" were collapsed into an *Endorsed as Relevant* category. As recommended by Polit et al. (2007), two item-level indices were calculated:

1) Item-level CVI (I-CVI) defined as the proportion of experts who endorsed an item as relevant, was calculated for all items in the scale.
2) Modified kappa (k^*) statistic was calculated using the formula provided by Polit et al. (2007) where $k^* = (\text{I-CVI} - p_c)/(1-p_c)$, and $p_c = [N!/A!(N-A)!]*0.5^N$ and N = total number of experts, and A = number of experts who endorsed the item as relevant for each item.

The modified kappa is calculated to address issues of chance agreement in the calculation of the I-CVI (Polit et al., 2007). Polit and colleagues (2007) suggest that for eight experts, items with a k^* greater than 0.74 are considered to have excellent agreement, items between 0.74 and 0.60 are considered to have good agreement, and items between 0.59 and 0.40 are considered fair. Items with $k^* < 0.40$ are considered poor. Items were considered for deletion if they had a k^* less than 0.60 (i.e., less than good agreement). Prior to deletion, items which were rated as relevant by five out of six caregivers but neither clinician were retained despite a k^* below the 0.60 cut-off. Endorsement of five out of six caregivers was selected in line with Polit and Beck's (2007) recommendations for the level of endorsement indicative of excellent relevance. These items were retained for inclusion in initial screening tool testing, as caregivers may observe a wider range of behaviours than clinicians given the extended time spent with the child in a range of settings. A third metric, the Scale-level CVI average (S-CVI/Ave), was calculated by taking the average of all the I-CVIs. For the S-CVI/Ave, a scale is considered to have acceptable validity if the S-CVI/Ave is greater than 0.80. The results of the expert panel review and refinements to the item pool will be discussed in subsequent sections.

Content Validation Results

Content Validity Indices Results

Of the initial 95 items, caregivers endorsed on average, 81.4% (range = 63.2% to 91.6%) of items as either quite relevant or very relevant, whereas clinicians endorsed 55.3% (range = 25.3% to 85.3%) of items. Table 3 displays the number of experts who endorsed each item and the I-CVI, and *k** for all items. As shown in Table 4.3, the I-CVI and *k** ranged from CVI = 0.25, and *k** = 0.16 for the item "Engages appropriately in group play" to CVI = 1.00 and *k** = 1.00 for 16 items such as "Acts emotionally and socially younger than peers" and "Often needs things repeated a number of times before they start to remember". The Scale-Level CVI average (S-CVI/Ave) was 0.749.

Item Pool Refinement

The items were refined through a two-stage process. Items with a low CVI and *k** were initially identified as candidates for removal. The research team then reviewed the items proposed for inclusion based on the CVI and *k** and the qualitative feedback to determine if additional items were removed or revised. Figure 4.2 shows the process of item pool development and subsequent refining, and Table 4.3 provides further details regarding the changes made to items and justification for removals/inclusion.

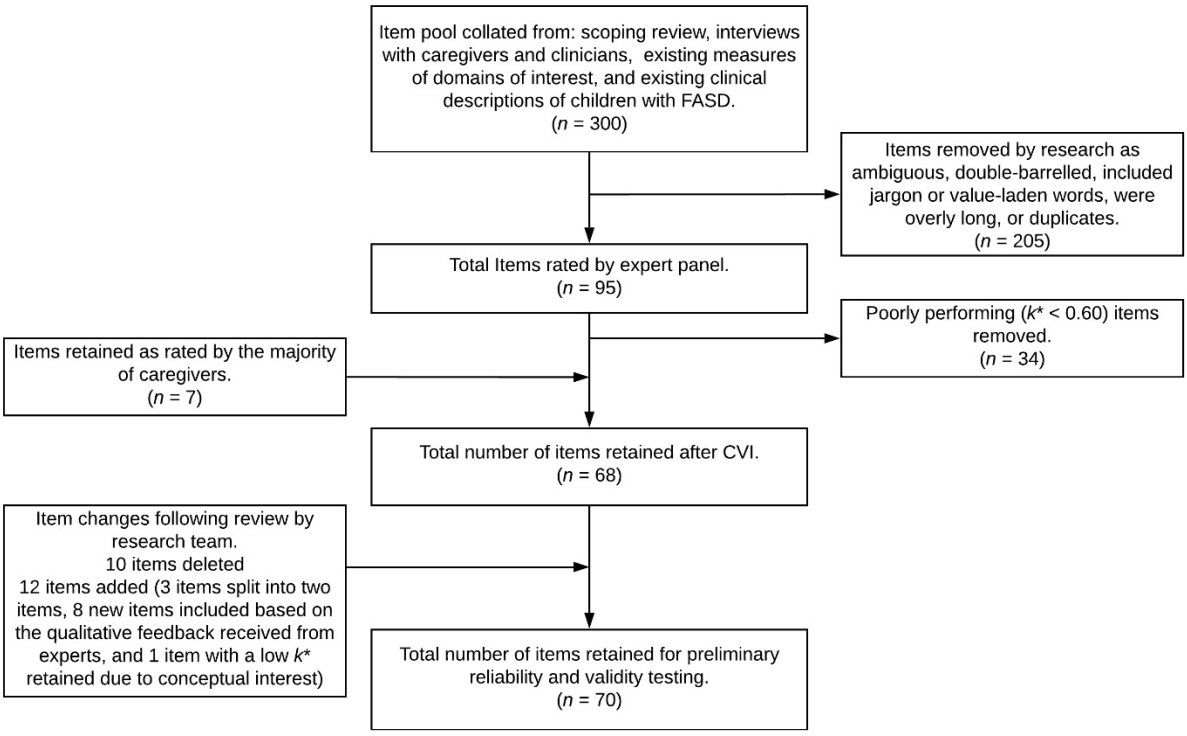


Figure 4.2. Numbers of items removed/retained through the item development process.

Refining based on CVI indices.

A total of 68 items made up of the 61 items endorsed by at least six of eight experts, and the seven items endorsed by five of six caregivers but neither clinician were retained based on the CVI and *k** indices. Sixty-one items were rated as relevant by at least six experts and were flagged for retention based on the *k**. Sixteen items were rated as relevant by all members of the expert panel. These highly-rated items included: “Chatty, but with little content”, “Easily led by peers”, “Makes the same mistakes repeatedly”, “ Acts emotionally or socially younger than peers”, and “Has difficulty understanding or anticipating the consequences of their actions”. A total of 34 items (18 adaptive functioning items and 16 self-regulation items) had a *k** below 0.60 and were flagged as candidates for item removal. No items from the cognitive functioning domain were identified for removal on the basis of CVI indices. Prior to deletion of the items

with a low *k**, items which were rated as relevant by five out of six caregivers but neither clinician were flagged. Seven items were found to meet this criterion (two adaptive behaviour items, five self-regulation items such as: “Resists going to bed at night”, “Exhibits wide mood swings”, “Has frequent meltdowns”, and “Runs about or climbs excessively in situations in which it is inappropriate” see Table 4.3). These items were reconsidered for inclusion in the tool despite having a low *k**. The S-CVI/Ave based on the 68 items was 0.835. The I-CVI ranged between 0.62 and 1.00, and *k** ranged between 0.52 and 1.00.

Further item list refinement.

Following the CVI process, the interim list of 68 items and the qualitative feedback from the experts were considered by the research team. Qualitative feedback was received on 26 items and related primarily to potential issues with reading and interpretation (e.g., requests for concepts to be clarified, splitting double-barrelled questions). The speech pathologist provided detailed feedback on the content relating to communication in the conceptual framework and more specific feedback on individual items. Several items were added considering this feedback and items such as “Unclear pronunciation of sounds/words” and “Makes errors with sentence structure (e.g., leaves words out, mixed up words, errors in tense)” were included. Qualitative comments were also made in relation to including items about children who are “low in mood and appear zoned out”; and including items about children who are “quieter or slower.” An item was added to capture this (“Appears to zone out or look lost in thought”). The research team reviewed each of the retained items to identify items that needed to be reworded. Potential duplication was also considered, and items were removed if they substantially overlapped with others. Decisions to remove overlapping items were made by consensus among the research team. During this review, the following changes were made:

- Seven items were deleted due to similarities with other items (e.g., “Needs repeated exposure to learn things properly” was deleted due to overlap with “Often needs information repeated a number of times before they start to learn and remember it”).
- Three items were split into two items each, adding three items (e.g., “Has difficulty making or keeping friends” was split into “Has trouble making friends” and “Has trouble keeping or maintain friends”).
- Eight new items were added, based on qualitative feedback from expert reviewers (e.g., “Appears to zone out or look lost in thought”, and “Appears to have difficulty applying learning across different settings”).
- One item which had been flagged for exclusion as having a low CVI was retained due to conceptual interest (e.g., “Behaviour is often inflexible”).

Table 4.3 details the items retained following the CVI phase, the changes made to the item and justifications, and the final items retained for inclusion in future testing (see Table 4.3 for the overall number of items at each stage). Lastly, all remaining items were revised to ensure item content fitted with the item stem: “Compared to other children their age, my child…”. Following final changes and review, a total of 70 items were included in the item selection for initial reliability, and validity testing of a new FASD screening tool.

Table 4.3

Item content, CVI, modified kappa, summary of item changes and final items, arranged by domain and sub-domain

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Adaptive functioning items					
Subdomain: Communication					
Chatty, but with little content	8	1.00	1.00	Edited for clarity	Is talkative, but with little depth or coherency
Uses simpler language than peers	7	0.88	0.88	Item split into two based on qualitative feedback.	Uses simpler words, and has less variety in words New Item: Uses simpler sentences than peers
Can't take a hint	6	0.75	0.72	Edited for clarity	Doesn't pick up on subtle social cues, such as body language, tone of voice, and facial expressions
Has difficulties understanding jokes	6	0.75	0.72	Exemplars added	Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)
Follows along with what is said/suggested, even if not true	6	0.75	0.72	Edited for clarity	Follows along with what is said/suggested, even if it is not true
Conversations with your child are often difficult for a listener to follow	5	0.63	0.52	Low CVI but retained due to 5/6 caregivers endorsing. Edited for consistency with item stem, exemplars added	Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)
Uses poor grammar in spoken sentences	5	0.63	0.52	Deleted due to low CVI	-
Lies when it would be easy to tell the truth	5	0.63	0.52	Deleted due to low CVI	-
Appears sincere when caught out in a lie	5	0.63	0.52	Deleted due to low CVI	-
Subdomain: Motor Skills					
Poor handwriting	7	0.88	0.88	Edited to clarify concept and added exemplars	Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)
Has problems with balance (e.g., struggles to stand on one leg)	6	0.75	0.72	Retained as is	Has problems with balance (e.g., struggles to stand on one leg)
Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	6	0.75	0.72	Retained as is	Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)
Difficulty handling, assembling, and manipulating small objects	4	0.5	0.31	Deleted due to low CVI	-

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Does not hold a crayon/pen/pencil properly	4	0.5	0.31	Deleted due to low CVI	-
Has difficulty throwing and catching a ball	4	0.5	0.31	Deleted due to low CVI	-
Has difficulty running fast	3	0.38	0.20	Deleted due to low CVI	-
Subdomain: Sensory Processing					
Does not dress appropriately for the weather	6	0.75	0.72	Retained as is	Does not dress appropriately for the weather
Decreased awareness of pain or temperature	5	0.63	0.52	Deleted due to low CVI	-
Subdomain: Sleep					
Resists going to bed at night	5	0.63	0.52	Low CVI but retained due to 5/6 caregivers endorsing Broadened to capture a range of sleep related behaviour	Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)
Is a light sleeper	3	0.38	0.20	Deleted due to low CVI	-
Subdomain: Social Skills					
Easily led by peers	8	1.00	1.00	Minor edit to fit item stem	Is easily led by peers
Has superficial friendships; is a social butterfly	7	0.88	0.88	Deleted on research team review as it was considered to overlap with the two items split from "Has difficulty making or keeping friends"	-
Has difficulty making or keeping friends	7	0.88	0.88	Split into two items	Has trouble making friends with similar age peers New item: Has trouble keeping, or maintain friendships
Is not aware of his or her actions on others	6	0.75	0.72	Added "appears" as inferring internal process	Appears unaware of the impact of his or her actions on others
Doesn't understand personal space	6	0.75	0.72	Removed the contraction, and added appears as inferring internal process	Does not appear to understand personal space
Has an air of competence	5	0.63	0.52	Retained following CVI process as rated relevant by 5 out of 6 caregivers, but deleted on research team review	
Indiscriminately friendly towards strangers	5	0.63	0.52	Deleted due to low CVI	-
Behaves in a way towards other children which they know annoys themselves	4	0.50	0.31	Deleted due to low CVI	-
Does not understand social cues (e.g., facial expressions, tone of voice, body language)	4	0.50	0.31	Deleted due to low CVI	-
Does not display remorse	4	0.50	0.31	Deleted due to low CVI	-
Acts on own needs or doesn't consider the needs of others	3	0.38	0.20	Deleted due to low CVI	-
Engages appropriately in group play	2	0.25	0.16	Deleted due to low CVI	-

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Subdomain: Young for Age					
Acts emotionally and socially younger than peers	8	1.00	1.00	Simplified the item, and added exemplars	Acts or behaves like a younger child (e.g., immaturity, emotionally younger)
Acts young for age	8	1.00	1.00	Deleted on research team review due to overlap with other items (Acts emotionally and socially younger than peers)	-
Able to engage more readily with younger peers, compared to same-age peers	8	1.00	1.00	Edited for clarity	Engages more readily with younger peers
Prefers the company of younger peers	8	1.00	1.00	Deleted on research team review due to overlap with other items (e.g., Able to engage more readily with younger peers, compared to same-age peers)	
Appears more capable than he or she really is	7	0.88	0.88	Deleted on research team review due to ambiguity	-
Appears not to have heard what you say and appears to ignore you	6	0.75	0.72	Edited for clarity	Appears to ignore people, or not listen to what is said
Cognitive functioning items					
Subdomain: Academic Ability					
Difficulties with abstract thinking	8	1.00	1.00	Exemplars added and edited for clarity	Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)
Struggles with complex maths problems (e.g., multiplication, word problems)	7	0.88	0.88	Retained as is	Struggles with complex maths problems (e.g., multiplication, word problems)
Struggles with literacy	7	0.88	0.88	Exemplars added	Struggles with literacy (e.g., reading comprehension, spelling, writing)
Has difficulties comprehending written information	7	0.88	0.88	Edited for clarity	Has difficulty understanding written information New item: has difficulties understanding verbal information
Is assigned schoolwork at a grade level below their current grade	7	0.88	0.88	Edited for clarity	Is assigned schoolwork at levels below their current year
Struggles with simple maths concepts (e.g., counting, arithmetic)	6	0.75	0.72	Retained as is	Struggles with simple maths concepts (e.g., counting, arithmetic)
Subdomain: General Cognitive Ability					
Has trouble learning and using the concept of time	8	1.00	1.00	Edited	Has difficulty understanding and applying the concept of time
Difficulty understanding and applying concepts such as now, or later, or days of the week	8	1.00	1.00	Edited for consistency with item stem	Has difficulty understanding and applying concepts such as now, later, before, or the days of the week

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Does not see the whole picture	6	0.75	0.72	Deleted on research team review due to ambiguity	-
Subdomain: Learning and Memory					
Often needs things repeated a number of times before they start to remember	8	1.00	1.00	Added "learn"	Often needs information repeated a number of times before they start to learn and remember it
Forgets things from one day to the next (for example, forgets school work that they knew the night before)	8	1.00	1.00	Added “appears” when inferring internal reasoning	Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)
Makes the same mistakes repeatedly	8	1.00	1.00	Edited for clarity	Repeats mistakes without seeming to realise or learn from previous attempts
Behaviour does not improve despite consistent consequences	8	1.00	1.00	Added “appear” when inferring internal reasoning, edited to fit with item stem	Does not appear to care about negative consequences to their behaviour
Needs repeated exposure to learning things properly	8	1.00	1.00	Deleted on research team review due to overlap with other items (e.g., Often needs information repeated a number of times before they start to learn and remember it)	-
Has trouble remembering to do things like school projects or chores	7	0.88	0.88	Edited for consistency with item stem	Has more trouble remembering to do school projects or chores
Struggles to recall recent information such as what just happened in a story, or what was just said to them	7	0.88	0.88	Edited to more clearly reflect the concept of interest	Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)
Often loses their lunch box, school work sheets, permission slips	7	0.88	0.88	Edited to make more general	Often loses their belongings (e.g., toys, lunch box, school work)
Has trouble completing tasks that have more than one step	7	0.88	0.88	Edited to simplify the item	Needs tasks broken down step by step
Struggles to remember activities that occurred during the day (e.g., what they did in the morning, what they ate)	7	0.88	0.88	Deleted on research team review due to overlap with other items (e.g., Forgets things from one day to the next (for example, forgets school work that they knew the night before))	-
Has trouble remembering things, even for a few minutes	6	0.75	0.72	Deleted on research team review due to overlap with other items (e.g., Struggles to recall recent information such as what just happened in a story, or what was just said to them)	-
Self-Regulation items					
Subdomain: Attention Regulation					
Needs help from an adult to stay on task	7	0.88	0.88	Minor edit to fit item stem	Needs more help from an adult to stay on task

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Is easily distracted by other things around them (e.g., other kids, things going on around them, background noise)	6	0.75	0.72	Removed exemplars	Is easily distracted by other things around them
Has trouble keeping track of what he/she is doing	5	0.63	0.52	Deleted due to low CVI	-
Has trouble remembering a list of tasks	5	0.63	0.52	Deleted due to low CVI	-
Subdomain: Behavioural Regulation					
Does things without first thinking about what could happen	8	1.00	1.00	Simplified and added “appears” when inferring internal reasoning	Appears to act without thinking
Has difficulties anticipating or understanding the consequences of their actions	8	1.00	1.00	Edited as double barrel question	Has difficulty anticipating the consequences of their actions
Is often "on the go" or acts as if "driven by a motor"	7	0.88	0.88	Retained as is	Is often "on the go" or acts as if "driven by a motor"
Has difficulty stopping an activity immediately upon being told to do so	7	0.88	0.88	Edited to make more general	Finds it difficult to stop doing something when asked to
Requires constant supervision	7	0.88	0.88	Edited for clarity	Requires constant or high levels of supervision
Does risky things	7	0.88	0.88	Exemplars added, and made more specific	Displays more risk-taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)
Destroys own property, or family’s property	7	0.88	0.88	Edited to make more general	Destroys belongings, either their own or others
Can’t be rushed between activities	7	0.88	0.88	Edited for clarity	Needs a lot of time to transition between activities
Talks excessively	6	0.75	0.72	Edited to include reference to other children	Talks much more than other children
Has difficulty playing or engaging in leisure activities quietly	6	0.75	0.72	Retained as is	Has difficulty playing or engaging in leisure activities quietly
Has difficulty awaiting turn	6	0.75	0.72	Edited to add reference to other children	Has more difficulty waiting for their turn than other children
Acts in a wilder way compared to other children when in a group (e.g., at a birthday party, during a group activity or on a school excursion)	6	0.75	0.72	Removed exemplars and clarified what was meant by “wilder”	Acts in a more hyperactive or excited way
Has difficulty handling exciting events	6	0.75	0.72	Edited for clarity	Is easily over-whelmed, or over-stimulated
Resists changes of routine	6	0.75	0.72	Revised to remove implication of intent	Has difficulty adapting to changes of routines
Replaces fact with fantasy	6	0.75	0.72	Edited as inferring internal processes	Appears to replace fact with fantasy
Has difficulty holding back his/her activity despite being told to do so	6	0.75	0.72	Deleted on research team review due to overlap with other items (e.g., Has difficulty stopping an activity immediately upon being told to do so)	-

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
Runs about or climbs excessively in situations in which it is inappropriate	5	0.63	0.52	Low CVI but retained due to 5/6 caregivers endorsing	Runs about or climbs excessively when it is inappropriate
Interrupts or intrudes on others (e.g., butts into conversations or games)	5	0.63	0.52	Low CVI but retained due to 5/6 caregivers endorsing	Interrupts or intrudes on others (e.g., butts into conversations or games)
Inflexible behaviour, for example gets stuck on an idea or object	5	0.63	0.52	Flagged for deletion due to low CVI but retained on research team review as was of interest to retain.	Behaviour is often inflexible (e.g., gets stuck on an idea or object)
Leaves seat in classroom or in other situations in which remaining seated is expected	5	0.63	0.52	Deleted due to low CVI	-
Doesn't attempt to hide evidence of misbehaviour (e.g., tells you when they have done something, they should know is wrong)	5	0.63	0.52	Deleted due to low CVI	-
Is overwhelmed in new situations or places	5	0.63	0.52	Deleted due to low CVI	-
Is aggressive towards siblings, peers or adults (e.g., parents, teachers)	5	0.63	0.52	Deleted due to low CVI	-
Is more difficult to manage in public than at home	4	0.50	0.31	Deleted due to low CVI	-
Subdomain: Emotional Regulation					
Has angry or tearful outbursts are intense but end suddenly	7	0.88	0.88	Angry or tearful collapsed into the category for emotional	Has emotional outbursts that end suddenly
Has outbursts for little reason	6	0.75	0.72	Added apparent when inferring internal reasoning	Has outbursts for little apparent reason
Is agitated if not getting their way	6	0.75	0.72	Revised for grammar and to fit item stem	Becomes agitated if they aren't getting their way
Exhibits wide mood swings	5	0.63	0.52	Low CVI, but retained due to 5/6 caregivers endorsing	Exhibits wide mood swings
Has frequent meltdowns	5	0.63	0.52	Low CVI, but retained due to 5/6 caregivers endorsing	Has frequent meltdowns
Has tantrums that last longer than would be expected from a child of that chronological age	4	0.50	0.31	Deleted due to low CVI	-
Often argues with adults	4	0.50	0.31	Deleted due to low CVI	-
Often quarrelsome or bossy with peers	4	0.50	0.31	Deleted due to low CVI	-
Has difficulty refraining from smiling or laughing in situations where it is inappropriate	3	0.38	0.20	Deleted due to low CVI	-
New items added					
					Unclear pronunciation of sounds/words
					Restarts sentences or words multiple times, as though they can't get the word out
					Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)

Item content	N experts endorsing	I-CVI	*k**	Item changes made[a]	Final items
					Appears to zone out or look lost in thought Appears to have difficulty applying learning across different settings Takes longer than other children to process information and respond Takes things which do not belong to them History of recurrent ear infections or hearing difficulties

Note. This table is arranged by over-arching domains, subdomains, and sorted by the total number of experts who rated an item as relevant.
I-CVI = Item-level Content Validity Index, *k** = modified kappa.
[a] Low $k^* < 0.60$

Discussion

This chapter presented the development, content validation and refinement of items for a screening tool intended to identify children with impairments consistent with those described among children with FASD. From an initial list of 300 items, 95 were reviewed by an expert review panel. Based on feedback received, a reduced list of 68 items were retained. The items retained in the screening tool had fair to excellent item-level I-CVI (I-CVI range = 0.63 to 1.00) and k^*(k^* range = 0.52 to 1.00) indices, and good whole scale content validity as measured by the S-CVI/Ave (S-CVI/Ave = 0.836) (Polit et al., 2007). After further refinement by the research team, a draft list of 70 items were retained for initial reliability and validity testing.

Items Retained

An intentionally broad set of items were retained at this early phase of scale development. Among the retained items were a set of items which, despite having a low I-CVI and a k^* in the "fair" range, were retained as they were rated as relevant by a majority of caregivers, but neither clinician. The retention of these items with lower item-level indices in turn impacted the S-CVI/Ave. It was, however, considered important to retain a broader set of items at the early stage of scale development given the previous literature review, interviews highlighting a wide range of impairments, and the considerable variation in behavioural presentation amongst children living with FASD. Further, as additional item refinement was planned during the initial testing, a broader set of items with adequate - if not exemplary - content validity was retained for initial testing.

In general, the items in the cognitive domain appeared to have greater proportions of endorsement than the self-regulation or adaptive functioning domains, and no items in the cognitive domain were removed due to low CVI. At the item level, items with the greatest CVI

related to appearing to behave like younger children, having difficulty understanding and anticipating the consequences of behaviour, impulsivity, and needing repeated exposure to learn new information. The scoping review (Chapter 2) identified consistent impairment in executive functioning which may under-pin the behavioural presentation of having difficulty anticipating consequences and behaving impulsively (Fuglestad et al., 2015). Items with low CVI reflected motor skills (e.g., difficulties throwing/catching a ball, and fine motor skills) and some aspects of social skills (e.g., not displaying remorse, acting on own needs and being quarrelsome or bossy). The items which were removed based on low CVI reflected domains which had mixed results in the interviews. For example, fine motors skills were a weakness for some, but not all children in the interviews, and being quarrelsome or bossy was reflected in relatively few interviews.

Differences Between Caregivers and Clinicians

Caregivers and clinicians differed with respect to the rates at which they endorsed items, with caregivers endorsing more items (81.4%) on average compared to clinicians (55.4%). The lower average appears to be driven by the low rates of endorsement by one clinician (paediatrician) who only rated 25.3% of items as relevant, whereas the second clinician (speech pathologist) endorsed 85.3% of the items. Caregivers showed less spread and had endorsement rates ranging from 63.1% to 91.6%. Caregiver endorsement of more items than clinicians may be a result of the length of time caregivers spend with their children, their knowledge of their children's behaviour, exposure to a greater range of behaviours, and observing children's behaviour in different contexts. The items retained as rated relevant by most caregivers but neither clinicians included items related to hyperactivity, emotional regulation, sleep, and communication skills.

It is also important to note that the interviews described in Chapter 3 and from which some of the domains and items were identified were conducted primarily with caregivers, and their perspectives may be more prominently reflected in the content of the item pool. A substantial body of literature has reviewed differences in child behaviour reports from the perspectives of different stakeholders such as parents, teachers, and the children themselves (Dirks, De Los Reyes, Briggs-Gowan, Cella, & Wakschlag, 2012). Dirks and colleagues (2012) suggest that differences may reflect situational specificity of behaviour. For example, sleep resistance is not likely to be assessed in a clinical setting unless explicitly raised (Ipsiroglu et al., 2013). Differences between parents or caregivers and stakeholders who interact with a wider number of children such as clinicians and teachers may also reflect access to a wider range of behaviour, may differ in their expectations of behaviour, and may also vary in their knowledge of behaviour (De Los Reyes, 2011). Clinicians also see children with various cognitive and behavioural presentation. The clinicians may interpret the questions to be what differences, if any, they see between children with FASD and those they see without neurodevelopmental or behavioural disorders, rather than in comparison to typically developing peers. The wording of the questionnaire completed by the expert panel, which asked their views on the proposed items for inclusion in a screening tool, may have been interpreted differently by experts. It is possible that the question was interpreted, in particular by the paediatrician who had low rates of endorsement, that they should identify the items that differ between children with FASD and other behavioural concerns that they see in their practice. Other respondents may have interpreted the question to reflect differences between children with FASD and typically developing peers.

It is important to note the professional backgrounds of the small number of clinicians who were part of the expert panel. Two clinicians participated in the research: one speech pathologist and one paediatrician. Differences in professional backgrounds were reflected in the feedback that experts provided on the proposed content and suggestions for edits and new items to be included, with the speech pathologist making several suggestions to capture difficulties with language in greater depth. As highlighted above, the clinicians also varied in the numbers of items that were endorsed as relevant. Based on a very limited sample of clinicians, it is difficult to draw conclusions as to whether the lower rates of endorsement are a consistent pattern, or an anomalous result. It is recommended that a multi-disciplinary team including psychologists, occupational therapists, and physiotherapists are involved in the diagnosis of FASD to ensure the full breadth of cognitive and behavioural functioning is assessed (Bower & Elliott, 2016).

Limitations

While this study met recommendations for the numbers of experts to recruit (Polit et al., 2007), the small number of clinicians is a limitation. Clinicians varied widely in their endorsements of the items which may reflect their differing clinical experience (e.g., speech pathologist and paediatrician). The low rate of clinician participation may impact upon the content validity of the tool. A small number of clinicians, representing an equally small number of professional backgrounds participated in the content validation study. Several professional groups who may frequently work with children with FASD did not participate in the content validation phase (e.g., occupational therapists, psychologists) despite attempts to increase recruitment as discussed above. Additional tool content which may be considered within scope of the assessments undertaken by these professional groups may not have been incorporated into the tool. Although professionals from these specialisms participated in the interviews and the

interviews were used to inform tool development, the lack of representation among the expert panel reviewing the tool may have limited their ability to identify additional items and ensure that the content of the tool reflect the difficulties displayed in their areas of specialization. This is evidenced in the additional items added based on the feedback received by the speech and language pathologist. For example, participation of occupational therapists in the content validation stage may have highlighted additional items to include which sought to further measure motor skills. To further enhance content validity, future research with a larger, more diverse pool of clinicians may identify additional items to further describe the difficulties experienced by children with FASD.

Difficulties were experienced in recruiting clinicians within Australia, which is representative of the relative infancy of the field with relative few experts at the time of the study. Clinicians with different areas of specialisation can add value to item development as demonstrated by the inclusion of additional communication items in the current study on the recommendation of the speech pathologist, however, with specialisation, agreement may be reduced between professionals. These groups could be contacted to increase the number of potential participants in any studies to further examine the content validity of the tool and clinician views.

The expert panel for this study was drawn from the same pool of participants who had participated in a previous phase of this research (See Chapter 3). The inclusion of those participants who had previously participated may limit the perspectives that are reflected in the items ultimately selected, as the same perspectives are reflected in both the generation of the items and the refinement. The caregiver with the lowest endorsement rates of items did not respond to the demographic questions, limiting the potential to examine possible reasons.

Further, while all clinicians participated in the previous qualitative phases, responses were only received from two of the five clinicians.

Conclusion

This chapter presented the development and content validation of a screening tool to assist in the early identification of children at-risk of having FASD. Through a multi-step process, a proposed screening tool was developed which demonstrated good face validity and content validity as rated by a panel of experts. Experts consistently identified behaviours such as appearing young for their age, difficulties understanding consequences, and impulsivity as relevant to the behavioural presentation of children with FASD. The next phase of this research will be to conduct initial reliability and validity tests to assess the psychometric properties of the proposed screening tool, and to provide initial estimates of the tool's reliability, validity, and accuracy in identifying children displaying impairments consistent with those described among children with FASD.

Chapter 5 Preliminary Reliability and Validity Study 1: Methods and Quantitative Item Analysis

The aim of this thesis is to develop a screening tool to identify children between the ages of four and 12 who display the cognitive and behavioural functioning consistent with having FASD. A preliminary 70-item screening tool- hereafter the Preliminary FASD Screening Tool (PFST)- representative of the behavioural and cognitive profile of children with FASD was developed based on the scoping review (Chapter 2) and interview findings (Chapter 3), and refined through review by an expert panel and the research team (Chapter 4) (Haynes et al., 1995; Streiner et al., 2015).

The preliminary reliability and validity study, as presented in this chapter, had two aims. Firstly, the study sought to further refine the PFST using quantitative item analysis. Quantitative item analysis refers to the variety of statistical procedures designed to ascertain the psychometric characteristics of individual items based on responses obtained from samples used in the process of test development (Urbina, 2004). The second aim of the study was to examine the reliability and validity of the PFST. The present chapter describes the methodology for the initial reliability and validity study, the sample, and the procedure and results of the quantitative item analysis process to further refine the content of the PFST. Reliability and validity indices calculations, which are dependent on the full scale, will be presented in Chapter 6.

Methodology

The preliminary reliability and validity study sought to further refine the PFST before examining the psychometric properties. The PFST is a proxy-reported measure of the behavioural and cognitive profile proposed to be characteristic of children aged four to 12 years living with FASD. A sample of caregivers raising a child in this age range who were either diagnosed with or

suspected of having FASD was recruited for the preliminary study. Participation was not restricted to caregivers of children with a confirmed diagnosis as many families raising children with FASD experience significant difficulties in accessing diagnostic services (Chasnoff et al., 2015). For the purposes of this study, the clinical group was defined as both those diagnosed as having FASD based on caregiver report and those suspected of having FASD based on caregivers' knowledge of PAE, the cognitive and behavioural functioning of their children, and any assessment conducted by health professionals to date. Further, it was considered that by making participation more inclusive, participants would be more likely to respond truthfully to questions about their child's diagnostic status. A second group of individuals who do not have the condition of interest are required for screening tool accuracy calculations (Evans et al., 2005; Streiner, 2003). Therefore, a community sample of caregivers of children aged four to 12 years was recruited.

Procedure and Participants

Ethics approval was received from University of South Australia Human Research Ethics Committee (UNISA HREC) (Protocol Number: 200987; Appendix F). Study advertisements were shared via social media and through mailing lists and newsletters of organisations supporting children and families living with FASD. Given that children with FASD are over-represented in out-of-home populations (Fuchs et al., 2010; Popova, Lange, Shield, et al., 2019), efforts were made to recruit foster and kinships carers. Calls for participation were distributed through the peak body for foster and kinship carer support organisations in South Australia. Further, to encourage participation among foster and kinship carers, a webinar about FASD and an advertisement for the research was delivered as part of regularly scheduled development activities for foster and kinship carers. The community sample was recruited using a convenience sample approach through social media and the personal networks of the research team. Study advertisements provided information

about the aims of the study and what participation would involve. Study advertisements (See Appendix G) directed participants to a study website containing more detailed information about the study, a link to a questionnaire, and links to information about FASD and support services for participants who may have experienced distress. All data were collected through Survey Monkey (https://www.surveymonkey.com/). The first page of the questionnaire outlined what participation would involve and the participants' rights. Participants indicated their consent via a check box before proceeding with the questionnaire (see Appendix H for the full questionnaire). Upon completion of the questionnaire, participants were redirected back to the study website, where they could download information about the support services and a copy of the information sheet for their records.

Participation was open to caregivers from Australia and New Zealand. While the questionnaire was initially only open to Australian participants, it was later opened to New Zealand participants with support received from the Australian and New Zealand FASD Clinical Network and Alcohol Healthwatch to adapt the distressed participant protocols and ensure participants who may have experienced distress could be supported. Participation by residents from other countries were excluded due to concerns raised by the UNISA HREC regarding the ability to provide support to participants should they experience distress. Potential participants from countries outside of Australia and New Zealand were excluded via eligibility questions at the outset of the questionnaire and directed to a page which thanked them for their interest in the study and outlined reasons why recruitment was limited to Australian and New Zealand participants.

Participants with multiple children aged between four and 12 were asked to keep their child whose birthday is next as the index child for the questionnaire. To aid participants

completing the survey, they were asked their child's name and were reminded throughout the questionnaire to keep this child in mind. Names were deleted from the data set prior to any analysis being conducted to preserve confidentiality.

Following completion of the questionnaire, participants were invited to sign up to a follow-up session wherein they would complete the FASD screening tool a second time for the purpose of test-retest reliability. The follow-up test-retest questionnaire was sent to participants via a nominated email address approximately two weeks after the first administration. Participants who did not complete the follow-up session were sent a single reminder email seven days after they had received the initial follow-up session email.

All participants completing the questionnaire were invited to enter a raffle for a $50 gift card. At the end of the questionnaire, participants could opt in to entering the raffle, at which point a second Survey Monkey window opened wherein participants could enter their details. This step was taken to ensure that raffle entries containing email addresses were kept separate from questionnaire responses for confidentiality purposes.

Materials

The online questionnaire was comprised of three major components: 1) Demographics and questions about lifestyle during pregnancy, 2) the PFST, and 3) Scales for convergent/divergent validation estimates. The demographic and lifestyle during pregnancy questions and PFST are described below. Full details of the scales included for convergent/divergent validation will be provided in Chapter 6.

Demographics and lifestyle during pregnancy questions.

The demographics component of the questionnaire included questions regarding age, ethnicity, education and employment. Participants were also asked questions about their child

including the child's age and gender, the participant's relationship to the child and any health concerns for their child. Participants who identified raising a child with FASD in the initial screening questions also completed a series of questions which sought information about the FASD diagnosis, including the diagnostic subcategory that was conferred, when the diagnosis was conferred and by whom, and what behaviours or signs led caregivers to pursue further investigation or diagnostic testing of their child. An option was provided for caregivers to indicate where a diagnosis was confirmed or provisional, pending further assessments.

The lifestyle during pregnancy questions sought information regarding each child's birthweight, gestational age, alcohol and tobacco exposure during pregnancy, and their birth mother's use of nutritional supplements in pregnancy. All participants were asked about the timing of any alcohol exposure in pregnancy, average number of drinks, how often drinks were consumed, episodes of binge drinking and whether more alcohol was consumed on special occasions (Muggli, Cook, O'Leary, Forster, & Halliday, 2015). Given that children with FASD may be over-represented among children removed from their birth parents (Popova, Lange, Shield, et al., 2019), the lifestyle during pregnancy questions were prefaced with a question about the source of information. This prefacing question was presented to participants who indicated that they were not the biological parent of the children. Participants could indicate their knowledge came from a personal relationship with and knowledge of the birth mother, conversations with the birth mother, court or medical documents, or other corroborating sources such as other family members.

Preliminary FASD Screening Tool.

Following the item development process outlined in Chapter 4, a pool of 70 items was retained for the quantitative item analysis. The content of the PFST reflects three over-arching

domains and sub-domains: adaptive behaviour (communication, motor skills, social skills), cognitive functioning (academic ability, general cognitive functioning, and learning and memory), and self-regulation (attention regulation, behavioural regulation, and emotional regulation). Participants rated items on a four-point scale, where 0 = never true, 1 = sometimes true, 2 = often true, and 3 = very often true. Potential scores on the PFST ranged from 0 to 210. All items were prefaced with the statement: "Compared to other children their age, my child…". Participants were instructed to keep their child's behaviour over the past six months in mind while completing the questionnaire.

Data Preparation

Questionnaire responses were exported from Survey Monkey and all data preparation and analysis was conducted using SPSS 26 (IBM Corp, 2019). Questionnaire responses were examined for completeness and eligibility. Cases were removed from analysis if they were missing a large proportion of the data (e.g., provided answers to demographic questions, but not on FASD screening tool items, $n = 47$) (Osborne, 2013). Of the 140 participants who commenced the survey, defined as clicking the link through from the study website to the information and consent sheet, 93 responses of sufficient completeness (90% or more of the demographics and PFST) were received. Although the questionnaire was set to limit participants to only completing the survey on one occasion, with the exception of test-retest reliability, three participants were found to have completed the questionnaire multiple times. Participants with duplicate responses were identified by the email addresses that were used to sign up to test-retest follow up session. One participant completed the questionnaire three times, and two participants completed the questionnaire twice, for a total of four duplicate responses. Only the first response from each participant was retained and second or subsequent responses were excluded from analysis.

While the target age range of the screening tool is children between the ages of four and 12, and this was made clear through recruitment materials and study information, no screening questions of child age were included. This was to encourage the accurate completion of questions relating to child age in the demographic section. Six responses were received from children outside the age range and were subsequently excluded from analysis. Following data cleaning, 83 responses were retained for analysis. Thirty-five participants completed a second administration of the PFST for test-retest reliability and analyses regarding test-retest reliability will be provided in the next chapter.

Prior to any analysis, several questions were recoded from free-text response boxes to dichotomous or scale variables. Free text responses for questions pertaining to gestational age and birth weight were recalculated to create a dichotomous variable indicating at-risk pregnancy outcomes of low birth weight and prematurity. Low birth weight was defined as a birth weight of less than 2,500 grams[11] (Australian Institute of Health and Welfare, 2018) and prematurity as any birth at less than 37 weeks[12] (Howson, Kinney, & Lawn, 2012). An "unknown" response was included where participants indicated a lack of knowledge of the child's birth weight or gestational age. Free-text questions for the length of time the child had been in the care of the participant, the age at which participant had first suspected FASD, and the age at which the diagnosis was conferred were transformed to provide a continuous variable for the length of time in years for each question.

[11] Where participants reported the weight in pounds and ounces, which were converted to grams using the following formula: $\text{Birthweight}_{grams}$ = ((pounds x 453.59237) + (ounces x 28.34952)).

12 Where participants reported gestational age in months, months were converted to weeks by multiplying by 4.345.

Data Analysis

Demographic data analysis.

Frequencies of responses to demographic questionnaires were examined. Chi-square and independent samples *t*-test analyses were used, where appropriate, to explore differences between the participants in the FASD and community samples on demographic variables. This included analysis of age, gender, relationship to their children, and number of children in the family.

Quantitative item analysis.

The quantitative item analysis (also called item reduction analysis) phase was conducted to ensure that "only parsimonious, functional, and internally consistent items are ultimately included" (Boateng et al., 2018, p. 149). The quantitative item analysis drew on established guidelines for item selection (Boateng et al., 2018; Clark & Watson, 1995; Streiner et al., 2015; Urbina, 2004). Common elements of quantitative item analysis[13] includes examination of the item discrimination, inter-item correlation and item-total correlation, and examination of missing cases (Boateng et al., 2018). Figure 5.1 shows the process for selecting items to be retained in the tool. An iterative process was adopted with the inter-item and item-total correlation steps repeated after any items were removed until no further items were removed.

[13] Other steps identified in quantitative item analysis including examination of the item difficulty index and distractor efficiency analysis were not conducted, as these analyses relate to items measuring knowledge of content (e.g., multiple choice items on a test of course content).

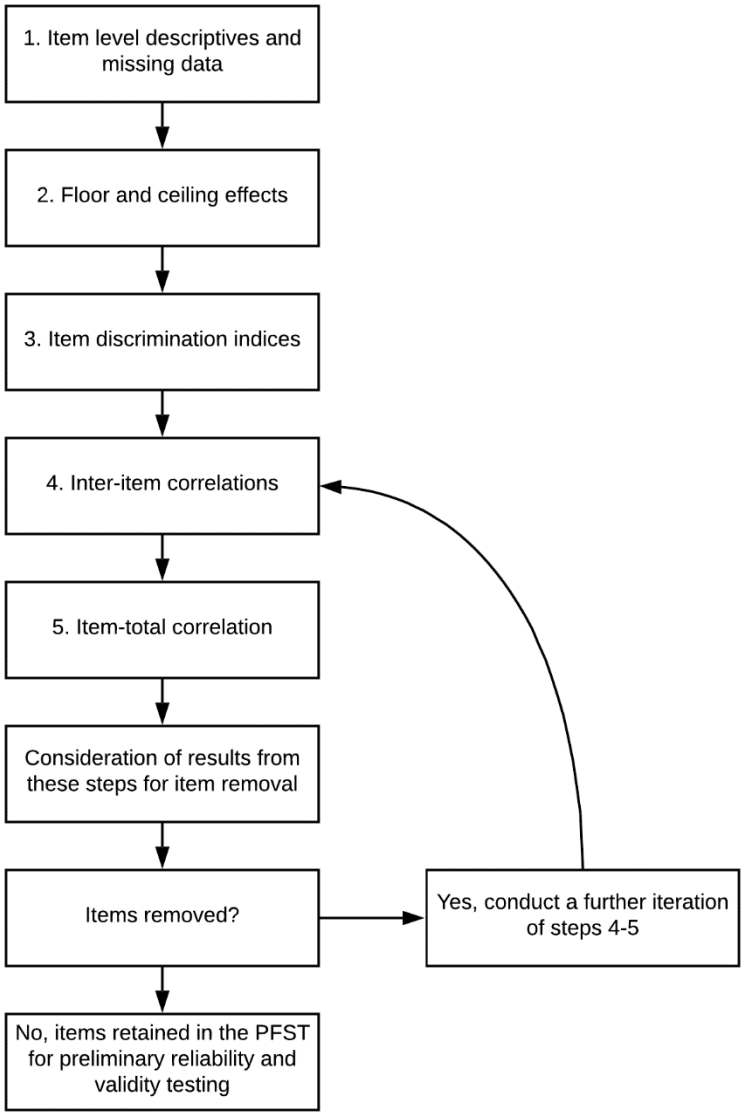


Figure 5.1. Item selection analysis.

Missing data.

For the quantitative item analysis phase, missing data were not replaced. Missing data patterns can provide useful information about items being considered for inclusion in the tool, and examination of the missing data forms part of the decision making around items to be retained (Streiner, Norman & Cairney, 2015). Patterns of missing data on the PFST were examined using

frequencies for the 83 participants retained in the sample for analysis. The percentage of missing data for each item was calculated to examine any potential bias, or items which were being skipped. Items skipped by large participant numbers were flagged as potential candidates for removal. Due to missing data at the item level, the sample size ranged from 71 for the item-total correlation analysis, as cases were excluded if they had any missing data, to 79-83 for the item discrimination and inter-item correlation analysis, with participants excluded from those analyses for which they had missing data.

Item level descriptive statistics.

Descriptive statistics (e.g., mean, standard deviation, proportion of missing data) were calculated for each item for the full sample, and the FASD and community samples separately. The rates of endorsement across the four response categories for each item of the screening tool were examined in the full sample. Items were flagged for removal if there were response categories which were endorsed by less than 5% of the full sample, or greater than 95% of the full sample (Streiner et al., 2015).

Item discrimination index.

Item discrimination refers to the extent to which items differentiate the subjects taking the test in terms of the characteristics that the test is intended to measure. Methods for calculating item discrimination include splitting the sample based on total scores (e.g., the top scoring 25% and the bottom scoring 25% of the sample) or to create groups based on an external criterion (e.g., membership in contrasted diagnostic groups) (Urbina, 2004). Given the goal of creating a screening tool to identify individuals who may have impairments consistent with those commonly described among children with FASD, the discrimination index was calculated using groups defined by diagnosis or suspicion of having FASD. In developing tests of complex and

multifaceted constructs, external criteria are preferred. Further, the use of external criteria is suggested to enhance test validity (Urbina, 2004). Appendix I shows the item discrimination indices calculated using both methods.

The item discrimination index was defined as the difference between the proportion of respondents who scored an item as either 2 = often true or 3 = very true in the FASD and community samples. The item discrimination index ranges from -1 to +1. Item discriminations above 0.4 are considered to be highly discriminating, and a cut-off of 0.4 was selected for retaining items in the PFST (Kortekaas et al., 2017). The item discrimination index was calculated using the following steps (Urbina, 2004):

1. The proportions of children in the diagnosed with or suspected of having FASD group who scored an item either 2= often true, or 3= very often true were calculated.
2. The proportions of children in the community sample who scored an item either 2= often true, or 3= very often true were calculated.
3. The community sample proportion was subtracted from the FASD group proportion.

Inter-item correlations and item total correlations.

A correlation matrix of all items was generated to identify item pairs or groups with high correlations, and to examine average inter-item correlations. Item pairs with correlation coefficients above 0.87 were flagged as potentially redundant, and these items were considered for removal (Pollard, Dixon, Dieppe, & Johnston, 2009). In the case of highly correlated pairs, where an item was considered for removal, the item with the lower item discrimination index was removed. Items with a low average inter-item correlation (< 0.15) were considered for removal (Clark & Watson, 1995). Lastly, corrected item-total correlations (the correlation of any item to

the total score, excluding that item) were calculated for all items. Items with low (< 0.3) item-total correlations were flagged for removal (Boateng et al., 2018; Streiner et al., 2013).

Results

Participant Demographics

Caregiver demographics.

Of the 83 participants, 42 caregivers reported they were raising a child either diagnosed with FASD or suspected of having FASD and 41 caregivers formed the community sample. Table 5.1 shows the caregiver demographic data. In total, 73 participants (32 FASD, 41 community sample) were from Australia, and 10 were from New Zealand (10 FASD, 0 community sample). The majority of survey responders were female regardless of the group (FASD = 92.86%, Community = 95.12%). Caregivers' ages ranged from 27 to 68 years, and caregivers of children with FASD (Mean = 46.27, SD = 8.66, Range = 27 to 68) were significantly older than the community sample (Mean = 41.15, SD = 5.28, Range = 32 to 57) (t (66.43) = 3.219, p = .002).

The caregivers of children with FASD and the community sample differed by both their level of education and employment status. Among the FASD sample the proportion of caregivers with undergraduate qualifications was 26.19% and 17% of the group had a post-graduate qualification. In the community sample, 85.37% reported having a postgraduate qualification. The majority of caregivers of children with FASD reported to either be engaged in full time or part time work (54.77%), and a proportion of caregivers of children with FASD reported either being unemployed (16.67%) or receiving a carer's pension (16.67%). The majority of caregivers in the community sample were engaged in part time or full-time work (92.68%).

Children with FASD were more likely to be in the care of others than their biological parents: 92.86% of caregivers of children with FASD indicated that they were not the biological

parents of the children in the study, compared to 9.76% of the community group. Further, children with FASD showed a wider range in the age at which they had been placed into the care of their current caregivers.

The majority of caregivers, across groups, reported they were raising more than one child, and caregivers of children with FASD reported significantly larger family sizes (FASD Mean number of children = 2.67, community sample = 2, $\chi^2 = 9.453$, $p = .022$). Additionally, 13 caregivers of children with FASD (30.95%) reported they suspected more than one child in their care had FASD. Of these, 10 caregivers reported that two children in their care either had FASD or were suspected of having FASD, and three caregivers reported that they had three children in their care with diagnosed or suspected FASD.

Table 5.1

Caregiver demographic characteristics

		FASD sample (n = 42)		Community sample (n = 41)		
		Mean	(SD)	Mean	(SD)	t-test
Age		46.27	8.660	41.15	5.284	3.201 ($p < .05$)
		n	%	n	%	Chi square
Gender	Female	39	92.86	39	95.12	0.00 ($p = 1.00$)
	Male	2	4.76	2	4.88	
	Missing	1	2.38	0	0.00	
Employment status	Employed full time	6	14.29	12	29.27	[1]
	Employed part time	17	40.48	26	63.41	
	Carers' pension	7	16.67	0	0.00	
	Retired	1	2.38	1	2.44	
	Studying	4	9.52	1	2.44	
	Unemployed	7	16.67	0	0.00	
Education	Less than year 12	5	11.90	0	0.00	[1]
	Completed secondary school, or equivalent	4	9.52	0	0.00	
	Vocational qualification	4	9.52	1	2.44	
	Diploma, or associate degree	11	26.19	0	0.00	
	Undergraduate qualification	11	26.19	5	12.20	
	Post graduate qualification	7	16.67	35	85.37	
Number of children	1	7	16.67	9	21.95	9.453 ($p = .022$)[2]
	2	16	38.10	24	58.54	
	3	9	21.43	7	17.07	
	4 or more	10	23.81	1	2.44	
Relationship to children	Biological parent	3	7.14	37	90.24	57.39 ($p < .001$)[3]
	Step-parent	2	4.76	0	0.00	
	Foster parent	17	40.48	2	4.88	
	Relative/kin	6	14.29	2	4.88	
	Adoptive parent	8	19.05	0	0.00	
	Legal guardian	6	14.29	0	0.00	
Age placed into care (FASD = 39, Community = 4)	Birth	15	38.46	3	75.00	[1]
	1	5	12.82	0	0.00	
	2	7	17.95	0	0.00	
	3	2	5.13	0	0.00	
	4 or older		23.08	0	0.00	
	Missing	1	2.56	1	25.00	
Country	Australia	32	76.19	41	100	11.10 ($p < .001$)
	New Zealand	10	23.81	0	0.00	

Note. [1] Chi square was not analysed on employment or education as the assumptions for Chi square tests were violated. Attempts were made to collapse the categories meaningfully to allow the analysis to be conducted but the violations of assumptions remained. [2] Number of children was assessed as a continuous variable and collapsed into categories for comparison across groups. [3] Chi square calculated by collapsing categories into biological (FASD = 3, community sample = 37) and caregiver other than biological parent (FASD = 39, community sample = 4).

Child demographics.

Table 5.2 shows the demographic details of the 83 index children. Children were, on average, 7.72 years old (SD = 2.62). Children with FASD (Mean age = 8.07 years) and the community sample (Mean age = 7.37 year) did not significantly differ in age ($t(82) = 1.23$, $p = .223$). While there were more male (55%) than female (42%) children in the sample,[14] the proportions of male and female children did not significantly differ across the FASD and community samples ($\chi^2 = 0.98$, $p = 1.00$). A large proportion of caregivers of children with FASD reported that they did not know if their child was born prematurely (Unknown = 13, 30.95%) or at low birth weight (Unknown = 14, 33.33%). Among the subsample that reported knowing this information, children with FASD were more likely to have been born prematurely (FASD = 30.7% premature, community sample = 5.2%; $\chi^2 = 8.14$, $p < .005$), or at a low birth weight (FASD = 37.5%, community sample = 2.5%; $\chi^2 = 14.30$., $p < .001$).

Of the 42 participants who indicated their child either had a diagnosis of FASD, or was suspected of having FASD, 32 (76.19%) children were reported to have a confirmed diagnosis. Of the 10 (23.81%) children without a confirmed diagnosis, three children had received some testing and were being monitored until being assessed again when they are older. The remaining seven children were reported to be either undergoing assessments or were waiting for service availability. For those with a confirmed diagnosis, the average age at diagnosis was 6.36 years old (SD = 2.14). Half of the caregivers of children with FASD reported their children had received a diagnosis of FAS, or FASD with three sentinel facial features (analogous under Australian diagnostic guidelines), and 40.63% reported a diagnosis of FASD with fewer than three sentinel

[14] One child was reported to identify as non-binary, hence the percentages of male and female children do not total 100%.

facial features. Two caregivers reported receiving a diagnosis of neurodevelopment disorder: alcohol exposed, and one caregiver reported they were unsure of their child's diagnosis. The majority of caregivers in the FASD group reporting a confirmed diagnosis indicated the diagnosis was made by a multi-disciplinary team (59.38%). When caregivers were asked whether their children had any health concerns, caregivers from the community sample reported one child to have dyslexia and one child was undergoing assessments related to a suspected autism diagnosis. Other health conditions reported related to physical health (e.g., asthma $n = 2$, and childhood heart disease $n = 1$).

Table 5.2

Child demographics

	FASD sample (n = 42)		Community sample (n = 41)		
	Mean	(SD)	Mean	(SD)	t-test
Age	8.07	2.54	7.37	2.69	1.23, (p = .223)
Age at diagnosis (n = 30, children with a confirmed diagnosis)	6.36	2.14	-	-	
	n	%	n	%	Chi square
Gender					
Female	18	42.86	17	41.46	0.98, (*p* > .05)
Male	23	54.76	23	56.10	
Non-binary	0	0.00	1	2.44	
Diagnostic status					
Confirmed	32	76.19	-	-	
Pending	10	23.81	-	-	
Diagnostic category (n = 32, those with a confirmed diagnosis)					
FASD with 3 Sentinel Facial Features, or FAS	16	50.00	-	-	
FASD with fewer than 3 Sentinel Facial Features,	13	40.63	-	-	
Unsure	1	3.13	-	-	
Other	2	6.33	-	-	
How diagnosis conferred (n = 32, those with a confirmed diagnosis)					
Multi-disciplinary team	19	59.38	-	-	
Paediatrician and psychologist	3	9.38	-	-	
Paediatrician coordinating multiple assessments	2	6.25	-	-	
Single clinician	8	25.00	-	-	
Prematurity[1]					
Premature (less than 37 weeks)	8	19.05	2	4.88	8.14, (*p* < .005)
Full term	18	42.86	38	92.68	
Unknown	13	30.95	1	2.44	
Missing	3	7.14	0	0.00	
Low birth weight[1]					
Less than or equal 2,500 grams	9	21.43	1	2.44	14.70, (*p* < .001)
More than 2,500 grams	15	35.71	40	97.56	
Unknown	14	33.33	0	0.00	
Missing	4	9.50	0	0.00	

Note. FAS = Fetal alcohol syndrome, FASD = Fetal Alcohol Spectrum Disorder.
[1] Participants reporting unknown or missing gestational age or birth weight were excluded from Chi square analyses.

Lifestyle during pregnancy information.

Caregivers who were not the biological parents (*n* = 42; FASD = 38, community = 4) of the children were asked the source of lifestyle information. Caregivers reported this information to come from a variety of sources including medical and legal documents (66.7%), personal relationship with the birth mother (19.0%), or conversations with the birth mother (26.2%)[15]. The results of the lifestyle during pregnancy questions are presented in Table 5.3. The majority of caregivers of children with FASD reported frequent high levels of alcohol exposure (e.g., multiple binge occasions each week) throughout pregnancy. As shown in Table 5.3, 35% (*n* = 14) of the community sample reported consuming some alcohol during pregnancy. Of these 14 participants, 10 reported drinking less than once a month and at low levels (typically one or two drinks in a drinking session), and to have no episodes of binge drinking. Varying patterns of alcohol consumption were reported by the remaining four participants: two participants reported one or two drinks once a month, one participant reported 3-4 drinks less than monthly. The community sample largely reported alcohol consumption throughout the duration of the pregnancy. Of the 14 who reported alcohol consumption in pregnancy, eight reported they drank throughout pregnancy, one participant reported drinking only in the first trimester, four participants reported drinking in the second or third trimesters, and one participant reported alcohol consumption in the first and third trimester. Further, a very high proportion (87.5%) of caregivers of children with FASD reported their children were exposed to tobacco smoking during pregnancy. Very low levels of smoking during pregnancy were reported by the community sample.

[15] Proportions total to more than 100% as caregivers could selected multiple options.

Table 5.3

Lifestyle during pregnancy[1]

		FASD sample (n = 42)		Community sample (n = 41)	
		n	%	*N*	%
Alcohol consumption	Yes	37	88.10	14	34.15
	No[2]	1	2.38	26	63.41
	Missing	4	9.52	1	2.44
Trimesters during which alcohol was consumed (FASD *n* = 37, Community *n* = 14)	1	34	91.98	10	71.43
	2	34	91.98	11	78.57
	3	32	86.49	11	78.57
Frequency	Daily or almost daily	19	51.35	0	0.00
	2 or 3 times a week	6	16.22	0	0.00
	Once a week	4	10.81	1	7.14
	Once every 2 weeks	0	0.00	1	7.14
	Once a month	0	0.00	2	14.29
	Less than once a month	1	2.70	10	71.43
	Missing	7	18.92	0	0.0
Amount on a drinking day	10 or more	8	21.62	0	0.0
	7–9	3	8.11	0	0.0
	5–6	2	5.41	0	0.0
	3–4	3	8.11	1	7.14
	1–2	1	2.70	11	78.57
	Unknown	15	40.54	1	7.14
	Missing	5	13.51	1	7.14
Frequency of binge exposure (4+ drinks/session)	Daily or almost daily	15	40.54	0	0.00
	2 or 3 times a week	5	13.51	0	0.00
	Once a week	3	8.11	0	0.00
	Once every 2 weeks	1	2.70	0	0.00
	Once a month	1	2.70	0	0.00
	Less than once a month	0	0.00	1	7.14
	Never	3	8.11	11	78.57
	Missing	9	24.32	2	14.29
Did you ever drink more on a special occasion?	Yes	26	70.27	4	28.57
	No	3	8.11	10	71.42
	Missing	8	21.62	0	0.00
Did you use any tobacco products during this pregnancy?	Yes	35	83.33	3	7.32
	No	5	11.96	37	90.24
	Missing	2	4.76	1	2.44
Trimesters during which tobacco was consumed (FASD *n* = 35, Community *n* = 3)	First	34	97.14	3	100
	Second	34	97.14	2	66.66
	Third	34	97.14	2	66.66

		FASD sample (n = 42)		Community sample (n = 41)	
Did you attempt to cut down on smoking during this pregnancy?	Yes	5	14.29	1	33.33
	No	27	77.14	2	66.66
	Missing	3	8.57	0	0.00

Note. [1] The lifestyle during pregnancy data violated the assumptions of chi square and so analyses could not be conducted. [2] One participant who was raising a child with FASD reported no alcohol consumption in pregnancy. Upon inspection, it was shown that the child in this case had received a FASD diagnosis with three sentinel facial features and reported receiving the diagnosis from a multi-disciplinary team. Taking this into account, the case was retained in the FASD group.

Quantitative Item Analysis Results

The results of the iterative item selection process are presented below. The item level descriptive statistics and percentages of missing data are presented first before the quantitative item analysis iterations. Thirteen items were removed on the first iteration of the quantitative item analysis. The analysis was re-run on the remaining 57 items, and no further items were removed.

Missing data.

Fourteen items in the screening tool were missing at least one response. The maximum number of missing responses for any item was three (3.61% missing) missing responses for the item “Is assigned schoolwork at levels below their current year”. At the participant level, nine participants were missing data on one item, one participant was missing data on two items, one participant was missing data on three items, and one participant was missing data on six items. Closer inspection revealed that this participant was the carer of a four-year old child and skipped all school-work related questions. No items were removed based on the proportion of missing data. It is worth noting, however, that the five questions relating to schooling were most frequently missed, albeit by only two or three participants per item.

Item-level descriptive statistics.

Table 5.4 shows the mean, standard deviation, minimum, and maximum for each item for the full sample and across the FASD group and the community sample. Children in the FASD

group appeared to have higher mean scores for each item of the 70-item list, with an average mean score of 2.17 compared to 0.45 among the community sample. Thirteen items among the FASD group had a restricted range, such that the lowest options (not true) were not endorsed. Among the FASD group, the items with a restricted range included seven items reflecting the self-regulation domain, one adaptive behaviour domain, and five cognitive domains. Items included: "Takes longer than other children to process information and respond" and "Has difficulty anticipating the consequences of their actions". Similarly, nine items among the comparison group had a restricted range, such that highest ratings (i.e., very often true) were not endorsed. Among the comparison group the items with a restricted range included two adaptive functioning items (e.g., "Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)"), two cognitive items (e.g., "Repeats mistakes without seeming to realise or learn from previous attempts"), and five self-regulation items (e.g., "Needs a lot of time to transition between activities"). A restricted range in both the FASD group and the community sample such that were no responses on the lowest response option among children with FASD, and no responses on the highest response option among the community sample was noted on the following three items: "Often loses their belongings (e.g., toys, lunch box, school work)", "Has difficulty adapting to changes of routines" and "Is easily distracted by other things around them".

For each item, the proportion of endorsement for each point of the 4-point scale was examined. When considering the full sample, items were inspected for the proportion of endorsement. Based on the full sample, each of the four options and items that were endorsed by greater than 95%, or less than 5% in any category were flagged for removal. Proportions of endorsement for each item are shown in Table 5.5. No items had endorsement rates for any response option greater than 95%, or less than 5%.

Table 5.4

Item level descriptive statistics

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
Adaptive functioning																
Communication and language																
13 Appears to ignore people, or not listen to what is said	1.55	1	1.12	0	3	2.4	3	0.77	0	3	0.68	1	0.65	0	2	0
50 Talkative, but with little depth or coherency	1.19	1	1.25	0	3	2.12	2	1.02	0	3	0.24	0	0.58	0	3	0
51 Unclear pronunciation of sounds/words	1.07	1	1.08	0	3	1.69	1.5	1	0	3	0.44	0	0.74	0	3	0
52 Restarts sentences or words multiple times, as though they can't get the word out	1.17	1	1.16	0	3	1.8	2	1.12	0	3	0.51	0	0.78	0	3	1
53 Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	1.08	1	1.18	0	3	1.78	2	1.13	0	3	0.37	0	0.73	0	3	1
54 Takes longer than other children to process information and respond[1]	1.36	1	1.33	0	3	2.45	3	0.8	1	3	0.24	0	0.66	0	3	0
55 Uses simpler words, and has less variety in words (smaller vocabulary)	1.04	0	1.21	0	3	1.81	2	1.11	0	3	0.24	0	0.7	0	3	0
56 Uses simpler sentences than peers	1.07	0	1.27	0	3	1.88	2	1.15	0	3	0.24	0	0.73	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
57 Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	1.22	1	1.28	0	3	2.21	2.5	0.95	0	3	0.2	0	0.56	0	3	0
59 Follows along with what is said/suggested, even if it is not true	1.18	1	1.17	0	3	1.95	2	1.01	0	3	0.39	0	0.7	0	3	0
60 Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	1.16	1	1.19	0	3	2.02	2	0.98	0	3	0.27	0	0.59	0	3	0
70 History of recurrent ear infections or hearing difficulties	0.77	0	1.19	0	3	1.17	1	1.3	0	3	0.37	0	0.92	0	3	1
Motor skills																
64 Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)	0.7	0	1.03	0	3	1.17	1	1.19	0	3	0.22	0	0.52	0	2	0
65 Has problems with balance (e.g., struggles to stand on one leg)	0.6	0	0.99	0	3	1.02	1	1.12	0	3	0.17	0	0.59	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
66 Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	0.89	0	1.1	0	3	1.5	2	1.09	0	3	0.27	0	0.71	0	3	0
Sensory																
63 Does not dress appropriately for the weather	1.31	1	1.11	0	3	1.88	2	1.06	0	3	0.73	1	0.84	0	3	0
Sleep																
69 Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	1.34	1	1.26	0	3	2.17	3	1.1	0	3	0.49	0	0.75	0	3	0
Social Skills																
42 Has trouble making friends with similar age peers	1.02	1	1.18	0	3	1.71	2	1.07	0	3	0.32	0	0.82	0	3	0
43 Has trouble keeping, or maintaining, friendships	1.06	1	1.22	0	3	1.88	2	1.06	0	3	0.22	0	0.69	0	3	0
44 Appears unaware of the impact of his or her actions on others	1.39	1	1.23	0	3	2.33	3	0.82	0	3	0.41	0	0.71	0	3	0
45 Does not appear to understand personal space	1.37	1	1.3	0	3	2.36	3	0.98	0	3	0.37	0	0.66	0	3	0
46 Is easily led by peers	1.59	1.82	1.24	0	3	2.54	3	0.81	0	3	0.63	0	0.8	0	3	1
49 Doesn’t pick up on subtle social cues, such as body	1.36	1	1.22	0	3	2.26	2.5	0.89	0	3	0.44	0	0.71	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
language, tone of voice, and facial expressions																
Young for age																
47 Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	1.36	1	1.26	0	3	2.26	3	0.91	0	3	0.44	0	0.81	0	3	0
48 Engages more readily with younger peers	1.17	1	1.2	0	3	2.02	2	0.98	0	3	0.29	0	0.64	0	3	0
Cognitive																
Academic ability																
14 Struggles with simple maths concepts (e.g., counting, arithmetic)	1.23	1	1.25	0	3	2.17	3	0.96	0	3	0.27	0	0.63	0	3	0
15 Struggles with complex maths concepts (e.g., multiplication, word problems)	1.58	2	1.29	0	3	2.54	3	0.81	0	3	0.55	0	0.85	0	3	2
16 Struggles with literacy (e.g., reading comprehension, spelling, writing)	1.37	1	1.22	0	3	2.22	2	0.88	0	3	0.5	0	0.85	0	3	2
17 Has difficulty understanding written information	1.44	1	1.21	0	3	2.29	2	0.81	0	3	0.53	0	0.85	0	3	2
18 Has difficulty understanding verbal information	1.2	1	1.21	0	3	2.14	2	0.87	0	3	0.24	0	0.58	0	3	0
19 Is assigned schoolwork at levels below their current year	1.2	1	1.21	0	3	2.15	3	1.08	0	3	0.28	0	0.82	0	3	3
General cognitive ability																

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
20 Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	1.12	1	1.21	0	3	2.05	2	0.95	0	3	0.15	0	0.53	0	3	2
61 Has difficulty understanding and applying the concept of time	1.37	1	1.28	0	3	2.36	3	0.82	1	3	0.37	0	0.77	0	3	0
62 Has difficulty understanding and applying concepts such as now, later, before, or the days of the week	1.22	1	1.21	0	3	2.02	2	0.95	0	3	0.39	0	0.83	0	3	0
67 Appears to zone out or look lost in thought	1.08	1	1.18	0	3	1.86	2	1.09	0	3	0.29	0	0.6	0	3	0
68 Appears to have difficulty applying learning across different settings	1.23	1	1.19	0	3	2.19	2	0.77	0	3	0.24	0	0.58	0	3	0
Learning and Memory																
21 Has more trouble remembering to do school projects or chores	1.11	1	1.22	0	3	2.57	3	0.74	0	3	0.48	0	0.78	0	3	1
22 Often needs information repeated a number of times before they start to learn and remember it	1.55	1.5	1.3	0	3	2.81	3	0.45	1	3	0.59	0	0.77	0	3	0
23 Appears to forget things from one day to	1.45	1	1.35	0	3	2.6	3	0.73	0	3	0.27	0	0.63	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
the next (e.g., forgets school work or rules that they knew the day before)																
24 Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	1.3	1	1.3	0	3	2.38	3	0.82	0	3	0.2	0	0.56	0	3	0
25 Often loses their belongings (e.g., toys, lunch box, school work)	1.31	1	1.16	0	3	2.22	2	0.82	1	3	0.37	0	0.54	0	2	1
27 Has difficulty anticipating the consequences of their actions	1.57	1	1.22	0	3	2.55	3	0.71	1	3	0.56	0	0.71	0	3	0
28 Repeats mistakes without seeming to realise or learn from previous attempts	1.54	1	1.33	0	3	2.69	3	0.68	0	3	0.37	0	0.58	0	2	0
29 Does not appear to care about negative consequences to their behaviour	1.43	1	1.31	0	3	2.45	3	0.86	0	3	0.39	0	0.74	0	3	0
41 Needs tasks broken down step by step	1.55	2	1.28	0	3	2.6	3	0.63	1	3	0.49	0	0.81	0	3	0
Self-regulation																
Attention regulation																
39 Is easily distracted by other things around them	1.59	2	1.19	0	3	2.57	3	0.63	1	3	0.59	0	0.67	0	2	0
40 Needs more help from an adult to stay on task	1.64	2	1.32	0	3	2.71	3	0.64	0	3	0.54	0	0.84	0	3	0
Behavioural regulation																
01 Talks much more than other children	1.57	1	1.06	0	3	1.86	2	1.16	0	3	1.27	1	0.87	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
02 Has difficulty playing or engaging in leisure activities quietly	1.46	1	1.2	0	3	2.29	3	0.89	0	3	0.63	0	0.84	0	3	1
03 Has more difficulty waiting for their turn than other children	1.37	1	1.2	0	3	2.26	2	0.77	1	3	0.46	0	0.81	0	3	0
04 Is often "on the go" or acts as if "driven by a motor"	1.57	1	1.22	0	3	2.43	3	0.89	0	3	0.68	1	0.82	0	3	0
05 Finds it difficult to stop doing something when asked to	1.75	2	1.08	0	3	2.57	3	0.67	1	3	0.9	1	0.7	0	3	0
06 Appears to act without thinking	1.65	1	1.17	0	3	2.61	3	0.67	1	3	0.68	1	0.69	0	3	1
07 Acts in a more hyperactive or excited way	1.59	1	1.24	0	3	2.57	3	0.8	0	3	0.59	1	0.67	0	3	0
08 Requires constant or high levels of supervision	1.59	1	1.24	0	3	2.45	3	0.77	1	3	0.32	0	0.69	0	3	0
09 Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	1.4	1	1.3	0	3	2.17	3	1.08	0	3	0.41	0	0.87	0	3	0
10 Behaviour is often inflexible, (e.g., gets “stuck” on an idea or object)	1.3	1	1.31	0	3	2.33	3	0.85	0	3	0.73	1	0.81	0	3	0
11 Runs about or climbs excessively when it is inappropriate	1.54	1	1.15	0	3	2.1	2	1.01	0	3	0.29	0	0.68	0	3	0
12 Interrupts or intrudes on others (e.g., butts into	1.2	1	1.25	0	3	2.43	3	0.83	0	3	0.98	1	0.79	0	3	0

	Full sample					FASD sample					Community sample					
Items	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Mean	Mdn.	Std Dev	Min	Max	Missing
conversations or games)																
26 Takes things which do not belong to them	1.29	1	1.16	0	3	1.48	1	1.09	0	3	0.27	0	0.5	0	2	0
34 Needs a lot of time to transition between activities	1.34	1	1.17	0	3	2.19	2	0.86	0	3	0.46	0	0.71	0	2	0
35 Is easily over-whelmed, or over-stimulated	1.55	2	1.27	0	3	2.55	3	0.74	0	3	0.54	0	0.81	0	3	0
37 Has difficulty adapting to changes of routines	1.41	1	1.16	0	3	2.31	2	0.75	1	3	0.49	0	0.68	0	2	0
38 Destroys belongings, either their own or others	0.93	0.5	1.12	0	3	1.6	1.5	1.15	0	3	0.23	0	0.48	0	2	1
58 Appears to replace fact with fantasy	1.13	1	1.23	0	3	2	2	1.04	0	3	0.24	0	0.62	0	3	0
Emotional regulation																
30 Has outbursts for little apparent reason	1.58	1	1.13	0	3	2.36	3	0.82	0	3	0.78	1	0.79	0	3	0
31 Has frequent meltdowns	1.43	1	1.2	0	3	2.24	2	0.85	0	3	0.61	0	0.92	0	3	0
32 Exhibits wide mood swings	1.33	1	1.17	0	3	2.02	2	1.02	0	3	0.61	0	0.83	0	3	0
33 Has emotional outbursts that end suddenly	1.2	1	1.13	0	3	1.9	2	1.03	0	3	0.49	0	0.71	0	3	0
36 Becomes agitated if they aren't getting their way	1.76	2	1.22	0	3	2.64	3	0.66	1	3	0.85	1	0.96	0	3	0

Note. Item numbers indicate the original order in which items were presented to participants in the questionnaire. For presentation in the table, items were re-arranged by the domains that they sought to measure. The item number has been retained to facilitate cross-referencing between items as appropriate.

Table 5.5

Proportions of endorsement of each response option

	Proportion of endorsement (%)			
	Never	Sometimes	Often	Very Often
Adaptive functioning				
Communication				
13 Appears to ignore people, or not listen to what is said	21.69	28.92	21.69	27.71
50 Talkative, but with little depth or coherency	44.58	16.87	13.25	25.30
51 Unclear pronunciation of sounds/words	37.35	34.94	10.84	16.87
52 Restarts sentences or words multiple times, as though they can't get the word out	39.02	26.83	13.41	20.73
53 Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	46.34	19.51	14.63	19.51
54 Takes longer than other children to process information and respond	42.17	13.25	10.84	33.73
55 Uses simpler words, and has less variety in words (smaller vocabulary)	50.60	15.66	13.25	20.48
56 Uses simpler sentences than peers	51.81	13.25	10.84	24.10
57 Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	45.78	13.25	14.46	26.51
59 Follows along with what is said/suggested, even if it is not true	39.76	22.89	16.87	20.48
60 Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	40.96	25.30	10.84	22.89
70 History of recurrent ear infections or hearing difficulties	65.85	9.76	6.10	18.29
Motor Skills				
64 Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)	61.45	18.07	9.64	10.84
65 Has problems with balance (e.g., struggles to stand on one leg)	66.27	16.87	7.23	9.64
66 Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	54.22	14.46	19.28	12.05
Sensory				
63 Does not dress appropriately for the weather	28.92	32.53	16.87	21.69
Sleep				
69 Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	37.35	21.69	10.84	30.12
Social Skills				
42 Has trouble making friends with similar age peers	48.19	20.48	12.05	19.28
43 Has trouble keeping, or maintaining, friendships	48.19	20.48	8.43	22.89
44 Appears unaware of the impact of his or her actions on others	34.94	19.28	18.07	27.71
45 Does not appear to understand personal space	39.76	15.66	12.05	32.53
46 Is easily led by peers	28.05	21.95	13.41	36.59

	Proportion of endorsement (%)			
	Never	Sometimes	Often	Very Often
49 Doesn't pick up on subtle social cues, such as body language, tone of voice, and facial expressions	34.94	20.48	18.07	26.51
Young for age				
47 Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	36.14	21.69	12.05	30.12
48 Engages more readily with younger peers	40.96	24.10	12.05	22.89
Cognitive				
Academic Ability				
14 Struggles with simple maths concepts (e.g., counting, arithmetic)	40.96	22.89	8.43	27.71
15 Struggles with complex maths concepts (e.g., multiplication, word problems)	32.10	18.52	11.11	38.27
16 Struggles with literacy (e.g., reading comprehension, spelling, writing)	34.57	20.99	17.28	27.16
17 Has difficulty understanding written information	33.33	18.52	20.99	27.16
18 Has difficulty understanding verbal information	40.96	20.48	15.66	22.89
19 Is assigned schoolwork at levels below their current year	48.75	12.50	7.50	31.25
General Cognitive Ability				
20 Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	46.91	17.28	13.58	22.22
61 Has difficulty understanding and applying the concept of time	37.35	19.28	12.05	31.33
62 Has difficulty understanding and applying concepts such as now, later, before, or the days of the week	39.76	22.89	13.25	24.10
67 Appears to zone out or look lost in thought	44.58	22.89	12.05	20.48
68 Appears to have difficulty applying learning across different settings	40.96	15.66	22.89	20.48
Learning and Memory				
21 Has more trouble remembering to do school projects or chores	32.93	17.07	12.20	37.80
22 Often needs information repeated a number of times before they start to learn and remember it	27.71	16.87	12.05	43.37
23 Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)	40.96	10.84	10.84	37.35
24 Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	43.37	13.25	13.25	30.12
25 Often loses their belongings (e.g., toys, lunch box, school work)	32.93	28.05	15.85	23.17
27 Has difficulty anticipating the consequences of their actions	26.51	25.30	13.25	34.94
28 Repeats mistakes without seeming to realise or learn from previous attempts	34.94	15.66	9.64	39.76
29 Does not appear to care about negative consequences to their behaviour	36.14	20.48	7.23	36.14
41 Needs tasks broken down step by step	32.53	15.66	15.66	36.14
Self-regulation				
Attention				
39 Is easily distracted by other things around them	25.30	22.89	19.28	32.53
40 Needs more help from an adult to stay on task	32.53	13.25	12.05	42.17
Behavioural Regulation				

	Proportion of endorsement (%)			
	Never	Sometimes	Often	Very Often
01 Talks much more than other children	15.66	39.76	16.87	27.71
02 Has difficulty playing or engaging in leisure activities quietly	26.83	30.49	10.98	31.71
03 Has more difficulty waiting for their turn than other children	33.73	20.48	20.48	25.30
04 Is often "on the go" or acts as if "driven by a motor"	26.51	25.30	13.25	34.94
05 Finds it difficult to stop doing something when asked to	13.25	33.73	18.07	34.94
06 Appears to act without thinking	20.73	30.49	12.20	36.59
07 Acts in a more hyperactive or excited way	25.30	28.92	7.23	38.55
08 Requires constant or high levels of supervision	38.55	15.66	13.25	32.53
09 Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	44.58	10.84	14.46	30.12
10 Behaviour is often inflexible, (e.g., gets “stuck” on an idea or object)	22.89	30.12	16.87	30.12
11 Runs about or climbs excessively when it is inappropriate	44.58	14.46	16.87	24.10
12 Interrupts or intrudes on others (e.g., butts into conversations or games)	14.46	33.73	18.07	33.73
26 Takes things which do not belong to them	46.99	31.33	8.43	13.25
34 Needs a lot of time to transition between activities	33.73	21.69	21.69	22.89
35 Is easily over-whelmed, or over-stimulated	31.33	18.07	14.46	36.14
37 Has difficulty adapting to changes of routines	30.12	22.89	22.89	24.10
38 Destroys belongings, either their own or others	50.00	23.17	10.98	15.85
58 Appears to replace fact with fantasy	46.99	14.46	16.87	21.69
Emotional Regulation				
30 Has outbursts for little apparent reason	20.48	31.33	18.07	30.12
31 Has frequent meltdowns	31.33	21.69	19.28	27.71
32 Exhibits wide mood swings	31.33	30.12	13.25	25.30
33 Has emotional outbursts that end suddenly	34.94	30.12	14.46	20.48
36 Becomes agitated if they aren’t getting their way	21.69	22.89	13.25	42.17

Note. Item numbers indicate the original order in which items were presented to participants in the questionnaire. For presentation in the table, items were re-arranged by the domains that they sought to measure. Number has been retained to facilitate cross-referencing between items as appropriate.

Item selection results: First iteration.

As shown in Table 5.6, 13 items were removed in the first iteration. Items removed are highlighted in bold with the justification included in the final column. Items were removed for low discrimination (n = 5, e.g., “Has problems with balance”, “Takes things which do not belong to them”) or displaying high correlations with another item/s (n = 6 e.g., “Has frequent meltdowns” was removed due to the high correlation with “Has outbursts for little apparent reason”). Upon inspection, two additional items were identified which reflected the potential secondary impacts of FASD, rather than the behavioural or cognitive manifestation and were removed (i.e., “assigned grade work below their level” and “requires constant supervision”). No items had a low item-total correlation (< .30) or low average inter-item correlation (< .15). In total, 13 items were removed. Four items were removed from the adaptive functioning domain reflecting motor skills (n = 2), and communication (n = 2). Four items were removed from the cognitive functioning domain reflecting academic achievement (n = 3) and learning and memory (n = 1). Five items from the self-regulation domain reflecting behavioural regulation (n = 3), emotional regulation (n = 1), and attention regulation (n = 1) were also removed.

Table 5.6

Qualitative item analysis results, first iteration

Item	Item Discrimination (*n* = 79 – 83)	Average inter-item correlation (*n* = 79 – 83)	Item-total correlation (*n* = 71)	Reason for removal. *Blank if question was retained.*
Adaptive functioning				
Communication				
13. Appears to ignore people, or not listen to what is said	.783	.686	.867	
57. Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	.761	.677	.874	
54. Takes longer than other children to process information and respond	.736	.738	.937	
50. Talkative, but with little depth or coherency	.714	.628	.79	
60. Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	.618	.692	.857	
59. Follows along with what is said/suggested, even if it is not true	.593	.602	.734	
53. Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	.585	.597	.75	
56. Uses simpler words, and has less variety in words (smaller vocabulary)	.57	.601	.758	
55. Uses simpler sentences than peers	**.546**	**.604**	**.76**	Removed. High inter-item correlation with item 56 "Uses simpler words, and has less variety in words (smaller vocabulary). Better discrimination noted for item 56.
52. Restarts sentences or words multiple times, as though they can't get the word out	.537	.558	.709	
51. Unclear pronunciation of sounds/words	.451	.534	.692	
70. History of recurrent ear infections or hearing difficulties	**.293**	**.357**	**.458**	Removed. Low discrimination.
Motor Skills				
66. Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	.523	.541	.716	

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)	Reason for removal. *Blank if question was retained.*
64. Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)	**.308**	**.474**	**.633**	Removed. Low discrimination.
65. Has problems with balance (e.g., struggles to stand on one leg)	**.237**	**.473**	**.63**	Removed.Low discrimination.
63. Does not dress appropriately for the weather	.473	.555	.707	
Sleep				
69. Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	.713	.542	.733	
Social Skills				
46. Is easily led by peers	.805	.688	.860	
45. Does not appear to understand personal space	.785	.690	.871	
44. Appears unaware of the impact of his or her actions on others	.76	.719	.901	
49. Doesn't pick up on subtle social cues, such as body language, tone of voice, and facial expressions	.736	.731	.913	
43. Has trouble keeping, or maintaining, friendships	.523	.657	.831	
42. Has trouble making friends with similar age peers	.474	.611	.779	
Young for age				
47. Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	.641	.704	.899	
48. Engages more readily with younger peers	.594	.677	.867	
Cognitive Ability				
Academic Achievement				
15. Struggles with complex maths concepts (e.g., multiplication, word problems)	.729	.620	.802	
18. Has difficulty understanding verbal information	.714	.709	.900	
17. Has difficulty understanding written information	.704	.658	.858	
16. Struggles with literacy (e.g., reading comprehension, spelling, writing)	**.631**	**.623**	**.826**	Removed. High inter-item correlation with item 17 "Has difficulty understanding written information" and item 16 displays lower item discrimination.
19. Is assigned schoolwork at levels below their current year	**.625**	**.640**	**.827**	Removed. Appears to be an impact question.
14. Struggles with simple maths concepts (e.g., counting, arithmetic)	.618	.640	.837	

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)	Reason for removal. *Blank if question was retained.*
General Cognitive Ability				
68. Appears to have difficulty applying learning across different settings	.809	.730	.924	
61. Has difficulty understanding and applying the concept of time	.713	.707	.883	
20. Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	.658	.729	.904	
67. Appears to zone out or look lost in thought	.595	.625	.800	
62. Has difficulty understanding and applying concepts such as now, later, before, or the days of the week	**.593**	**.685**	**.859**	Removed. Displays high inter-item correlation with item 61 "Has difficulty understanding and applying the concept of time" and displays poorer discrimination and psychometric properties. Is less clear than item 61.
Learning and Memory				
28. Repeats mistakes without seeming to realise or learn from previous attempts	.880	.733	.925	
23. Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)	.856	.742	.936	
22. Often needs information repeated a number of times before they start to learn and remember it	.854	.740	.930	
41. Needs tasks broken down step by step	.831	.738	.925	
21. Has more trouble remembering to do school projects or chores	**.83**	**.706**	**.894**	Removed. High correlations with several other items (Items 22, 23) that all appear to measure memory and have greater item discrimination.
24. Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	.809	.729	.915	
27. Has difficulty anticipating the consequences of their actions	.808	.709	.896	
29. Does not appear to care about negative consequences to their behaviour	.761	.699	.880	
25. Often loses their belongings (e.g., toys, lunch box, school work)	.732	.664	.842	
Self-regulation				
Attention Regulation				

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)	Reason for removal. *Blank if question was retained.*
39. Is easily distracted by other things around them	.831	.737	.915	
40. Needs more help from an adult to stay on task	**.830**	**.736**	**.927**	Removed. Displays high correlations with a number of items (8, 39, 41). Item 39 and 41 displayed higher item discrimination. Item 40 removed.
Behavioural Regulation				
06. Appears to act without thinking	.829	.733	.910	
07. Acts in a more hyperactive or excited way	.808	.686	.864	
35. Is easily over-whelmed, or over-stimulated	.807	.720	.894	
08. Requires constant or high levels of supervision	**.760**	**.748**	**.931**	Removed. Appears to be an impact question.
05. Finds it difficult to stop doing something when asked to	.758	.699	.871	
37. Has difficulty adapting to changes of routines	.736	.720	.900	
03. Has more difficulty waiting for their turn than other children	.712	.677	.849	
04. Is often "on the go" or acts as if "driven by a motor"	.711	.616	.777	
09. Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	.688	.684	.878	
10. Behaviour is often inflexible, (e.g., gets “stuck” on an idea or object)	.688	.635	.801	
02. Has difficulty playing or engaging in leisure activities quietly	.687	.636	.777	
58. Appears to replace fact with fantasy	.666	.659	.820	
11. Runs about or climbs excessively when it is inappropriate	.665	.709	.899	
34. Needs a lot of time to transition between activities	.640	.692	.863	
12. Interrupts or intrudes on others (e.g., butts into conversations or games)	.638	.659	.816	
38. Destroys belongings, either their own or others	.475	.589	.736	
26. Takes things which do not belong to them	**.38**	**.544**	**.698**	Removed. Low discrimination.
01. Talks much more than other children	**.308**	**.239**	**.319**	Removed. Low discrimination
Emotional Regulation				
30. Has outbursts for little apparent reason	.711	.664	.821	
36. Becomes agitated if they aren’t getting their way	.710	.717	.894	
31. Has frequent meltdowns	**.639**	**.684**	**.853**	Removed. High inter-item correlation with item 30 and displays poorer psychometrics than item 31.
33. Has emotional outbursts that end suddenly	.546	.584	.735	

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)	Reason for removal. *Blank if question was retained.*
32. Exhibits wide mood swings	.521	.610	.764	

Note. Item numbers indicate the original order in which items were presented to participants in the questionnaire. For presentation in the table, items were re-arranged by the domains that they sought to measure. Number has been retained to facilitate cross-referencing between items as appropriate.

Item selection results: Second iteration.

The item-discrimination, inter-item correlations, and item-total correlations were re-run on the 57 items retained in the screening tool. No further items met the criteria for removal in the second iteration of the quantitative item analyses, as shown in Table 5.7. The 57 retained items made up the version of the PFST for testing the scale-level reliability, validity, and accuracy in differentiating children with FASD from the community sample (see Chapter 6).

Table 5.7

Qualitative item analysis results, second iteration

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)
Adaptive functioning			
Communication			
13. Appears to ignore people, or not listen to what is said	.783	.686	.867
57. Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	.761	.677	.874
54. Takes longer than other children to process information and respond	.736	.738	.937
50. Talkative, but with little depth or coherency	.714	.628	.79
60. Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	.618	.692	.857
59. Follows along with what is said/suggested, even if it is not true	.593	.602	.734
53. Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	.585	.597	.750
56. Uses simpler words, and has less variety in words (smaller vocabulary)	.570	.601	.758
52. Restarts sentences or words multiple times, as though they can't get the word out	.537	.558	.709
51. Unclear pronunciation of sounds/words	.451	.534	.692
Motor Skills			
66. Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	.523	.541	.716
63. Does not dress appropriately for the weather	.473	.555	.707
Sleep			
69. Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	.713	.542	.733
Social Skills			
46. Is easily led by peers	.805	.688	.860
45. Does not appear to understand personal space	.785	.690	.871
44. Appears unaware of the impact of his or her actions on others	.760	.719	.901
49. Doesn't pick up on subtle social cues, such as body language, tone of voice, and facial expressions	.736	.731	.913
43. Has trouble keeping, or maintaining, friendships	.523	.657	.831
42. Has trouble making friends with similar age peers	.474	.611	.779
Young for age			

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)
47. Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	.641	.704	.899
48. Engages more readily with younger peers	.594	.677	.867
Cognitive Ability			
Academic Achievement			
15. Struggles with complex maths concepts (e.g., multiplication, word problems)	.729	.620	.802
18. Has difficulty understanding verbal information	.714	.709	.900
17. Has difficulty understanding written information	.704	.658	.858
14. Struggles with simple maths concepts (e.g., counting, arithmetic)	.618	.640	.837
General Cognitive Ability			
68. Appears to have difficulty applying learning across different settings	.809	.730	.924
61. Has difficulty understanding and applying the concept of time	.713	.707	.883
20. Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	.658	.729	.904
67. Appears to zone out or look lost in thought	.595	.625	.800
Learning and Memory			
28. Repeats mistakes without seeming to realise or learn from previous attempts	.88	.733	.925
23. Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)	.856	.742	.936
22. Often needs information repeated a number of times before they start to learn and remember it	.854	.74	.93
41. Needs tasks broken down step by step	.831	.738	.925
24. Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	.809	.729	.915
27. Has difficulty anticipating the consequences of their actions	.808	.709	.896
29. Does not appear to care about negative consequences to their behaviour	.761	.699	.880
25. Often loses their belongings (e.g., toys, lunch box, school work)	.732	.664	.842
Self-regulation			
Attention Regulation			
39. Is easily distracted by other things around them	.831	.737	.915
Behavioural Regulation			
06. Appears to act without thinking	.829	.733	.91
07. Acts in a more hyperactive or excited way	.808	.686	.864
35. Is easily over-whelmed, or over-stimulated	.807	.720	.894
05. Finds it difficult to stop doing something when asked to	.758	.699	.871
37. Has difficulty adapting to changes of routines	.736	.720	.900

Item	Item Discrimination (n = 79 – 83)	Average inter-item correlation (n = 79 – 83)	Item-total correlation (n = 71)
03. Has more difficulty waiting for their turn than other children	.712	.677	.849
04. Is often "on the go" or acts as if "driven by a motor"	.711	.616	.777
09. Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	.688	.684	.878
10. Behaviour is often inflexible, (e.g., gets “stuck” on an idea or object)	.688	.635	.801
02. Has difficulty playing or engaging in leisure activities quietly	.687	.636	.777
58. Appears to replace fact with fantasy	.666	.659	.820
11. Runs about or climbs excessively when it is inappropriate	.665	.709	.899
34. Needs a lot of time to transition between activities	.640	.692	.863
12. Interrupts or intrudes on others (e.g., butts into conversations or games)	.638	.659	.816
38. Destroys belongings, either their own or others	.475	.589	.736
Emotional Regulation			
30. Has outbursts for little apparent reason	.711	.664	.821
36. Becomes agitated if they aren’t getting their way	.710	.717	.894
33. Has emotional outbursts that end suddenly	.546	.584	.735
32. Exhibits wide mood swings	.521	.610	.764

Discussion

The aim of this chapter was to refine the item list presented in Chapter 4 using quantitative items analysis and to develop the PFST for completion by caregivers of children aged four to 12 years old. Participants raising children with FASD were recruited through social media and mailing lists of organisations supporting individuals with FASD and the community sample was recruited using a convenience sampling approach. Data gathered in this study were used to reduce the 70-item pool to a 57-item screening tool. Decisions to remove items were guided by several analyses including the extent to which individual items discriminated between children with FASD and the community sample, the correlations between items and the total scale, and consideration of the content of the items themselves. Of the 57 items retained, 21 items reflected over-arching self-regulation, 16 reflected cognitive ability, and 20 reflected adaptive functioning. The retained items all demonstrated adequate psychometric properties and met the thresholds established for inclusion in the tool.

Items Removed or Retained in the PFST

The reasons for removal of items varied by the over-arching domain. Reflective of the high levels of relevance noted for the cognitive domain items in chapter 4, the cognitive items all strongly discriminated between children with FASD and the community sample. Four items were removed from the cognitive domain. Three items were removed due to high correlations and overlap with other items. Two items “Is assigned schoolwork below their current grade” and “Requires constant supervision” were removed as these items appeared to be examining the outcomes of impairments associated with FASD, rather than direct impairments. No items were removed from the cognitive domain as poor discriminators. Further, of the three missing responses to item 19, two caregivers who skipped this question were caregivers of children who

were four and five years old, and the question may have been skipped as it may not have been applicable (i.e., the children were not at school yet). This may suggest that there are insufficient response options (e.g., needing a not applicable option), or rewording the item is needed to include kindergarten and preschool.

In the adaptive functioning domain, four items were removed, largely due to poor discrimination between the FASD and community sample. Interestingly, items examining motor skills have largely been removed from the tool, with the exception of one item ("Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)") which examines the acquisition of motor skills and may reflect memory/learning processes in addition to motor skills. In the self-regulation domain, two items were removed due to a low discrimination index ("Takes things which do not belong to them" and "Talks much more than other children"). A further two items were removed as they significantly overlapped with another item, and a final item was removed as it was believed to represent the secondary impacts associated with FASD rather than a behavioural or cognitive feature. These items included "Is assigned schoolwork at levels below their current year" and "Requires constant or high levels of supervision".

Characteristics of the Two Samples

There was a considerable difference between the FASD group and the community sample with respect to caregiver education and living situation. The community sample reported having predominantly postgraduate education qualifications and were largely the biological parents of children in the study, whereas the FASD sample had a wider spread of levels of education, in line

with the general population[16] and were for the most part, carers other than biological carers. While the caregivers of children with FASD were significantly older than their community sample peers (FASD mean age = 46.27(8.66), community mean age = 41.15(5.84)), this is consistent with the older age of carer populations (Siminski, Chalmers, & McHugh, 2005)

Differences between the FASD and community samples in parental education may be contributing to the differences noted on the mean and median scores of the PFST items (Table 5.4) and the level of discrimination between the FASD group and community sample (Table 5.6). Parental education has been shown to be a protective factor for child development (Duncan & Magnuson, 2012; Walker et al., 2011), and with such high levels of caregiver education, children in the community sample recruited in this study may have less difficulty in the domains studied than a representative sample of the general population. This may further increase differences between groups that were expected on the basis of having a FASD diagnosis or not. Alternatively, differences that may result from the differences in caregiver education may have been incorrectly assumed to reflect the FASD diagnosis.

Recruitment was conducted through organisations set up to support children and families living with FASD (the FASD group), and through convenience sampling using social media and personal networks (community sample). These recruitment strategies may themselves have contributed to the sampling bias in several ways. Firstly, the audience of the FASD support networks including the National Organisation for Fetal Alcohol Spectrum Disorders may be comprised of two groups: families living with FASD and professionals (e.g., health care workers, allied health workers, teachers, social workers, researchers) who have a professional interest in

[16] The Australian Bureau of Statistics reports that for females between 15 and 74, 10.4% have postgraduate qualifications (either postgraduate degree, or graduate diploma/certificate) and 20.4% have undergraduate qualifications (Australian Bureau of Statistics, 2019). These figures would include those too young to have completed undergraduate qualifications, as well as the older generation where the proportions are likely to be lower.

FASD and "follow" these networks to keep abreast of developments in the field. This audience of professionals may bias the level of education noted among the community sample. Previous research has suggested that convenience sampling strategies among community populations tends to find higher levels of education among volunteers participating in research than the general population (Ganguli, Lytle, Reynolds, & Dodge, 1998; Pruchno et al., 2008).

The majority of the caregivers raising children with FASD reported they were not the biological parents of children (92.9%), compared to 9.7% of the community sample reporting they were not the biological parents. This is important for several reasons including the accuracy of information about PAE as identified by non-biological caregivers, comorbid exposure to trauma in addition to PAE, and placement instability. Previous studies have reported high proportions of caregivers raising children with FASD to be non-biological carers. As stated in Chapter 2, previous studies examining children with FASD have frequently reported a higher portion of children with FASD to be in the care of carers other than biological parents. Among the 27 studies to report this information, the proportion of children no longer in the care of their birth parents ranged between 32% and 100%. Recent research from the UK, USA and Canada also report a similarly high proportion of children no longer in the care of their birth parents when recruiting families living with FASD through support organisations for children living with FASD. In this sample of over 250 caregivers, approximately 2% were birth parents (Price, 2019; Reid & Moritz, 2019). Further, children with FASD are thought to be over-represented amongst children who are removed from the care of their parents (Fuchs et al., 2010; Popova, Lange, Shield, et al., 2019). Foster and kinship carers who are raising a child living with FASD may be more likely to engage with support groups, such as those that supported recruitment in this study, compared with biological parents raising children with FASD. Owing to the stigma associated

with FASD (Choate & Badry, 2019), and possibility of on-going substance use concern, biological families of children with FASD may be considered a hard to reach population.

It is not known whether children in the FASD sample who were not living with their biological parents were the subject of child protection or family court orders or were living in informal arrangements and whether these children had experienced abuse or neglect in childhood, in addition to PAE. Such experiences may significantly impact on their cognitive and behavioural development (Dinkler et al., 2017; McLaughlin et al., 2017; Nemeroff, 2016; Teicher & Samson, 2016). The placement history of children prior to their current placement was also not assessed, and they may have been through several placement changes prior to their current placement. Placement breakdown is associated with impaired behavioural functioning (Delfabbro, King, & Barber, 2010; Rubin, O'Reilly, Luan, & Localio, 2007) and may have impacted scores in the FASD group. A number of caregivers of children with FASD reported that children had been placed into their care when they were two years old or older, and information is not available on placement stability prior to this date. Length of time spent in stable home is an important predictor of better outcomes among children with FASD (Streissguth et al., 2004). Given that many caregivers were not the birth parents of children with FASD, this may also influence the accuracy to which they can speak to the quantity of alcohol exposure and other prenatal or post-natal exposures. Similarly, many children in the FASD group were reported to have exposed to tobacco during pregnancy. Exposure to tobacco during pregnancy has been shown to negatively impact children's cognitive and behavioural development (Cornelius & Day, 2009).

The community sample contained both a number of children who were prenatally exposed to alcohol (n = 14, 35.15%), and a small number of children with developmental

conditions (undergoing assessment related to suspected ASD diagnosis n = 1, 2.44%, and dyslexia n = 1, 2.44%). Removal of these individuals from the community sample may artificially inflate the specificity of the PFST, by presenting a more homogenised comparison group that could be described "wellest of the well" and inflating group differences (Kohn, Carpenter, & Newman, 2013, p. 1202). Further, given the goal of the thesis was to create a screening tool that could be applied in a universal setting, these children were retained in the community sample to better represent the samples in which such a tool may be applied.

There was a spectrum of alcohol exposure reported amongst the community sample. Caregivers in the community sample who reported alcohol use during pregnancy (*n* = 14, 35) were retained in the community sample for analysis. The majority (70) of caregivers in the community sample who reported alcohol use during pregnancy reported low levels of alcohol exposure: 1-2 drinks, less than monthly, and with no reports of binge drinking. The rate of alcohol consumption noted among the community sample in this study was slightly lower than the National Drug Strategy Household Survey (NDSHS) which reported that 56% of women continued to drink during pregnancy. However, the level of alcohol consumption reported in this sample was similar as the NDSHS reported that of pregnant women who drank during pregnancy, 78% reportedly drank monthly or less (Australian Institute of Health and Welfare, 2014). Given that the same sample was used for the initial scale-level reliability and validity testing, the limitations and potential explanatory mechanisms are further discussed in Chapter 6 and the over-arching discussion in Chapter 7.

Future Directions for Research

This preliminary study had a small sample size, and as a result, the quantitative item analysis was conservative. At this stage of screening tool development, it was the intention to be

overinclusive of items. While there may be some redundancy in the questionnaire as noted by the high item-total correlations and average inter-item correlations, there will be opportunities to further refine the questionnaire in subsequent studies with a larger pool of participants and using methods such as factor analysis. Factor analysis was not conducted in the current sample due to sample size concerns. For future testing, a three factor structure matching the three overarching domains identified in the interviews (Chapter 3), and consistent with the ND:PAE criteria (American Psychological Association, 2013) could be examined. As this was initial testing, further item reduction steps can be taken using factor analysis procedures with a larger study and with more representative community and FASD samples or with a more diverse comparison group (i.e., individuals with ADHD, a comparison group of children who have experienced trauma) to further refine the proposed tool. A further consideration for future research is how to address the age-related questions that were skipped by individuals whose children may have been too young to relate to the questions. Lastly, participants in the study were aware that they were undertaking a study about FASD and this may have introduced a potential response bias. Future research, particularly among the general population, may consider whether to incorporate an element of concealment into the methodology given the stigma surrounding FASD (Choate & Badry, 2019).

Conclusions

This chapter sought to further refine the content of the PFST through a qualitative item analysis process. After examination of the item discrimination, item total correlations, and inter-item correlations, 13 items were removed from the proposed content of the PFST. The 57 retained items covered a range of cognitive and behavioural functioning domains. The retained items demonstrated good psychometric properties, meeting the thresholds established for tool

inclusion. Several important limitations are noted, particularly the bias between the community comparison group and the children with FASD. In the following chapter, the reliability (internal consistency and test-retest reliability), validity (convergent/divergent validity, and differences across known groups) and screening accuracy of the 57-item PFST will be examined.

Chapter 6 Preliminary Reliability and Validity Study 2: Reliability and Validity

Chapters 2 - 5 outline the development of the Preliminary FASD Screening Tool (PFST), following recommended stages of scale development (Haynes et al., 1995; Streiner et al., 2015). The item pool was developed drawing from the results of a scoping review and interviews with caregivers of children with FASD and clinicians with experience in FASD diagnostic and assessment roles. The PFST was reviewed for content validity by an expert panel comprised again of caregivers and clinicians. The PFST showed an acceptable level of content validity (Chapter 4) and the individual items showed acceptable levels of reliability (average inter-item correlation and item-total correlation) and discriminated well between children with FASD and a community sample in a quantitative item analysis process (Chapter 5). In the present chapter, the reliability and validity of the 57-item PFST will be examined.

Following recommendations for the assessment of reliability and validity, reliability will be examined using both internal consistency (the extent to which the items on test are inter-correlated) and test-retest reliability (how stable the scores are over time) (Boateng et al., 2018; Clark & Watson, 1995). The construct validity of the PFST will be examined through a series of analysis. Firstly, scores on the PFST will be examined across demographic variables. The ability of a newly developed scale to differentiate across binary groups (i.e., differentiation by known groups) that are expected to differ based on theoretical or empirical knowledge of performance of the binary groups is considered an element of construct validity (Boateng et al., 2018). Children diagnosed with or suspected of having FASD can be expected to score higher on the PFST. Correlations between the PFST and measures of related constructs will be performed to examine the convergent and divergent validity of the PFST (Boateng et al., 2018). The

performance of the PFST in differentiating between children diagnosed with or suspected of having FASD and the community sample will be examined using Receiver Operating Characteristics (ROC) curves and by exploring the sensitivity and specificity of the PFST. Finally, the accuracy of the PFST in differentiating between these two groups will be compared to the Neurobehavioural Screening Tool (NST). The NST is a brief screening tool to identify children who display behavioural symptomatology consistent with having FASD.

Methodology

Participants for the reliability and validity testing of the tool were the same as described in Chapter 5. Participants were included if they had largely complete data on the PFST (> 90%). A total of 83 participants had complete PFST data, however, the sample with available data for scales included for validation varied as two participants did not complete all measures. Subsequently, the sample size varied across analyses. For analyses of the PFST (e.g., group comparisons and sensitivity/specificity analysis) all 83 participants were included. For analysis of the measures included for convergent/divergent validation involving the measures which participants did not complete, the sample size is reduced to 81. Further, there were 35 (FASD = 20, Community sample = 15) participants who participated in the follow up survey to determine the test-retest reliability of the PFST.

In addition to the demographic data collected as outlined in the previous chapter, the following measures were included for estimating the convergent and divergent validity of the PFST, the NST, the SDQ, the Childhood Executive Function Inventory (CHEXI), and the Brief Assessment Checklist for Children (BAC-C).

Measures

Preliminary FASD Screening Tool.

The PFST was developed to identify children who display cognitive and behavioural impairments consistent with having FASD, based on their cognitive and behavioural presentation. As detailed in the preceding chapters, the PFST contains 57 items, measuring self-regulation, adaptive and cognitive function domains. The PFST is scored on a four-point scale where 0 = Not true, 1 = Rarely true, 2 = Often true, 3 = Very often true.

Childhood Executive Function Inventory.

Children with FASD frequently experience difficulties in executive functioning (Kingdon, Cardoso, and McGrath (2015), Chapter 2), and executive functioning is considered a hallmark difficulty of children with FASD (Kodituwakku & Kodituwakku, 2014). The CHEXI measures similar content as the behavioural regulation, and cognitive ability domains of the PFST. As a result, a measure of behaviour and cognition associated with FASD would be expected to correlate highly with a measure of executive functioning. The CHEXI (Thorell & Nyberg, 2008) is a proxy-reported measure of executive functioning and was included in the present study to establish convergent validity between the PFST and an established measure of executive functioning. The CHEXI consists of 24-items, rated on a 5-point scale, which ranges from 1 = definitely not true to 5 = definitely true. It generates two subscale scores: working memory and inhibition (Catale, Meulemans, & Thorell, 2015; Thorell & Nyberg, 2008). Scores are calculated by summing relevant items, with higher scores indicating greater executive functioning difficulty. Questions include “Has difficulty coming up with a different way of solving a problem when he/she gets stuck” and “When asked to do several things, he/she only remembers the first or last”. Current study participants were asked to read each statement

carefully and indicate how true each statement was for their child. The CHEXI does not specify a timeframe (e.g., past six months) for considering a child's behaviour when caregivers are responding. Previous research has established good psychometric properties for the CHEXI, including internal consistency and test-retest reliability (Cronbach's α = .89, test-retest r =.87 for the working memory index, and Cronbach's α = .85, test-retest r =.74 for the inhibition index (Catale et al., 2015)). As a test of convergent validity, it was predicted that there would be a significant moderate to strong positive correlation between the PFST and the CHEXI Working Memory and Inhibition subscales.

Strengths and Difficulties Questionnaire.

Individuals with FASD frequently have poorer parent ratings on measures of behavioural problems (Tsang, Lucas, Olson, Pinto, & Elliott, 2016). The SDQ (Goodman, 1997) is a widely used measure of behavioural problems among children and adolescents and is frequently used as a screening tool to identify children who are at risk of behavioural or emotional problems. The SDQ has been used extensively in research, including amongst children prenatally exposed to alcohol to measure behavioural and emotional problems (Alvik, Aalen, & Lindemann, 2013; Niclasen, Andersen, Strandberg-Larsen, & Teasdale, 2014; Sayal et al., 2014). The SDQ comprises 25 items, scored on a 3-point scale, where 0 = not true, 1 = somewhat true and 2 = certainly true. The SDQ generates scores on five subscales: Emotional Problems, Conduct Problems, Hyperactivity, Peer Problems, and Prosocial, and a total problems scale generated by summing four of the subscales (Emotional Problems, Conduct Problems, Hyperactivity, and Peer Problems). Higher scores on the Emotional Problems, Conduct Problems, Hyperactivity, and Peer Problems subscales indicate greater behavioural problems, with higher scores on the prosocial scale indicating more prosocial behaviour and fewer behavioural problems. Example

items on the SDQ include “Often has temper tantrums” and “Kind to younger children”. In responding to the SDQ, participants are asked to keep their child’s behaviour over the last six months in mind. The SDQ has shown good psychometric properties in previous research with good internal consistency for the total problems scale (Cronbach’s $\alpha = .79$), and adequate internal consistency for the subscales (Emotional Problems $\alpha = .73$, Conduct Problems $\alpha = .64$, Hyperactivity $\alpha = .80$, Peer Problems $\alpha = .61$, Prosocial $\alpha = .75$) among an Australia sample. (Seward, Bayliss, Stallman, & Ohan, 2018). Further, the SDQ demonstrates good test-retest reliability (average correlation between test administrations $r = .76$) when completed by parents (Stone, Otten, Engels, Vermulst, & Janssens, 2010).

As a test of convergent validity, it was predicted that there would be a significant moderate to strong positive correlation between the PFST and the SDQ total problems and the Emotional Problems, Conduct Problems, Hyperactivity, Peer Problems subscales. Further, a moderate negative correlation was anticipated between the PFST and the SDQ Prosocial subscale.

Neurobehavioural Screening Tool.

The NST (Nash et al., 2006)) is a 10-item screening tool for FASD, adapted from the Child Behaviour Checklist (CBCL (Achenbach & Rescorla, 2001)). Items on the NST include “Has your child been seen or accused of or thought to have acted too young for his or her age” and “Has your child been seen or accused of or is thought to display acts of cruelty, bullying or meanness to others”. Items are dichotomously scored, as yes or no and respondents are asked to keep their child’s behaviour over the previous six months in mind. The NST shows variable sensitivity (range = 63% – 98%) and specificity (range = 42% – 100%) when differentiating children with FASD and typically developing controls (Breiner et al., 2013; LaFrance et al.,

2014; Nash et al., 2011; Nash et al., 2006). Further, accuracy of the NST in differentiating children with FASD from ADHD has been examined, with sensitivity reported to range between 81% and 89%, but variable sensitivity, ranging between 42% and 72%. (Nash et al., 2011; Nash et al., 2006). No other reliability or validity information appears to be available for the NST.

The NST is scored using an algorithm to identify potential FASD cases based on scoring patterns (Nash et al., 2011) The NST scoring has two algorithms. The first (hereafter NST-A) is intended to identify individuals with behaviour suggestive of FASD. The second (hereafter NST-B) is intended to differentiate between children who display behaviour suggestive of FASD without ADHD symptomatology. In addition to the scoring algorithms, total scores are calculated, as per LaFrance et al. (2014). The NST was included in the present study to enable comparison between the newly developed PFST and existing FASD-specific screening tools. It is anticipated that there will be a high positive correlation between the NST and the PFST. Further, accuracy rates (sensitivity, specificity, positive predictive power and negative predictive power) were compared across the NST and the PFST, to compare their accuracy in identifying children who display the cognitive and behavioural profile consistent with FASD.

Brief Assessment Checklist for Children.

Children with FASD are often reported to experience significant post-natal adversity including abuse, neglect and removal from biological parental care (Kambeitz et al., 2019). Experiences of early life trauma can impact upon children's cognitive and behavioural development (D'Andrea, Ford, Stolbach, Spinazzola, & Van Der Kolk, 2012; McLaughlin et al., 2017). The BAC-C was included to measure behaviours related to experiences of trauma as conceptually distinct from cognitive and behavioural functioning associated with FASD.

The BAC-C is a screening checklist developed to capture clinically significant attachment- and trauma- related mental health problems experienced by children and young people living in alternative care (e.g., foster, kinship and residential care) that are not adequately captured by the CBCL (Tarren-Sweeney, 2013). The BAC-C was developed from the Assessment Checklist for Children (Tarren-Sweeney, 2007). The BAC-C is comprised of 20 items, rated on a 3-point scale, where 0 = not true, 1 = partly true, and 2 = mostly true. Items include "Fears that you will reject him/her" and "Relates to strangers as if they were family". In answering the BAC-C, participants are asked to keep the child's behaviour over the last four to six months in mind. The 20 items on the BAC-C are summed to create a single score. Previous research with the BAC-C has found acceptable internal consistency (α =.89; Tarren-Sweeney, 2013). Test-retest reliability estimates for the BAC-C appear to be unavailable. Given the over-representation of children with FASD who are no longer in the care of their birth parents, the BAC-C was included as a measure of behaviour which may be more reflective of attachment- or trauma-related experiences. The PFST was expected to correlate moderately with the BAC-C.

Data Preparation

Missing data.

Prior to data analysis, the 57-items of the PFST and the measures included for convergent/divergent validation were examined for missing data (Osborne, 2013). Table 6.1 summarises the proportion of missing data, and the approach taken to missing data for each of the scales in the questionnaire. Two participants withdrew prior to completing the questionnaires for convergent/divergent validity and no data was available from these participants on several measures. These participants were excluded from analyses involving the scales for which they

had no data available, and as a result the sample size varied across the analysis[17]. Full details of how missing data was approached for each scale are presented in Appendix J.

Table 6.1

Missing data summary

Questionnaire	Participants with > 90% complete data	Level of additional missing data	Missing data approach
PFST	83	10 participants missing at least one question.	Missing questions replaced with individual mean
SDQ	83	4 participants missed one question	Missing data replaced with pro-rata scores, as per developer instructions (Youthinmind, n.d.)
CHEXI	82	2 participants skipped one question	Missing questions replaced with individual mean
BAC-C	81	None	-
NST	81	None	-
PFST Retest[2]	35	None	-

Note. Tests were presented sequentially, and 2 participants did not complete all measures. The total available sample ranged from 83 for the PFST and SDQ, to 81 for the BAC-C and NST.
PFST = Preliminary FASD Screening Tool, SDQ = Strengths and Difficulties Questionnaire, CHEXI = Childhood Executive Functioning Inventory, BAC-C = Brief Assessment Checklist for Children, NST = Neurobehavioural Screening Tool.

Checking included measure assumptions.

Scales were scored according to the instructions available from the developers. Prior to data analysis, scales included for convergent and divergent validation were examined against the assumptions of normality, linearity, and examined for potential outliers (Tabachnick & Fidell, 2013). Deviations from normality were noted across the PFST, the SDQ subscales and total score, the CHEXI working memory and inhibition indices, the NST and the BAC-C. Scales typically had a negative kurtosis (as shown in Appendix K). Non-parametric equivalent tests were selected for analyses. Correlations were conducted using Spearman's rank-order correlation and between-groups analyses were conducted using Mann-Whitney U tests, as appropriate.

[17] Two participants dropped out before completing all scales. (1. Not diagnosed with FASD, screen negative on PFST, 4 years old, Male. 2. diagnosed with FASD, screen positive on PFST, 12 years old, Male)

Data Analysis

Data analysis was conducted using SPSS (IBM Corp, 2019). Accuracy metrics were calculated using the MedCalc web-based calculators (https://www.medcalc.org/calc/diagnostic_test.php) (MedCalc, n.d.).

Internal consistency and reliability.

Internal consistency and test-retest reliability estimates were calculated for the 57-item PFST for 83 participants. Internal consistency was estimated using Cronbach's α, and the average inter-item correlations of the retained items (Clark & Watson, 1995). Cronbach's α above .70 are suggested to indicate fair internal consistency, and above .90 are suggested for good internal consistency (Streiner et al., 2015). An average inter-item correlation between 0.15 and 0.50 is suggested to demonstrate sound internal consistency. Average inter-items correlations above 0.50 can indicate that items too similarly, and suggest redundancy (Clark & Watson, 1995). Test-retest reliability for the PFST was calculated using Intra-Class Correlation with a two-way mixed effects model, with absolute agreement (Koo & Li, 2016). Values of the ICC estimates below 0.5 are considered poor, between 0.5 and 0.75 are considered adequate, between 0.75 and 0.9 are considered good and above 0.9 is indicative of excellent reliability (Koo & Li, 2016).

Differentiation by known groups.

The construct validity of the PFST was tested by examining scores on the PFST across known groups, and relationships with demographic variables (Boateng et al., 2018). Mean scores on the PFST were compared across children diagnosed with or suspected of having FASD and the community sample, and across genders. It was anticipated that children with FASD will score higher than the community sample on the PFST. Relationships between the PFST and age were

also examined using Spearman's rank-order correlation. Mean scores of children diagnosed with or suspected of having FASD and the community sample were compared on SDQ, NST, CHEXI, and the BAC-C, using Mann-Whitney U Tests. Effect sizes for the Mann-Whitney U were calculated using the formula $r = Z / \sqrt{N}$ (Field, 2013). Guidelines for effect sizes suggest that .5 is a large effect, .3 is a moderate effect, and .1 is a small effect (Field, 2013).

Convergent and divergent validation.

Correlations between the PFST total score, SDQ total problems and Emotional Problems, Conduct Problems, Hyperactivity, Peer Problems, and Prosocial subscales, CHEXI Working Memory and Inhibition subscales, the NST total score, and the BAC-C total score were examined using Spearman's rank-order correlation.

Accuracy of the PFST in identifying children diagnosed with, or suspected of having FASD, and comparison to the NST

The accuracy of the PFST in identifying children diagnosed with or suspected of having FASD was examined using Receiver Operating Characteristics (ROC) curves, and an examination of sensitivity and specificity, and the positive and negative predictive power of the tool (Streiner, 2003; Streiner & Cairney, 2007). The examination of the accuracy of the PFST involved a multi-step process of creating a ROC curve to examine the sensitivity and specificity at each possible cut point (the score on the test used to designate a positive or negative screening outcome), selecting an optimal cut point based on the accuracy of the PFST and the intended use, and examining the accuracy of the PFST using the sensitivity, specificity, likelihood ratios, and positive and negative predictive power (Streiner, 2003).

Analysis was conducted using the SPSS ROC Curve Analysis function to generate a ROC Curve, calculate the Area Under the Curve (AUC), and calculate the sensitivity and specificity at

each possible cut-point on the PFST. The decision on which cut-point to select was guided by two indices: Youden's *J* and the Minimum Distance (Carter, Pan, Rai, & Galandiuk, 2016).

In addition to being guided by the Youden's *J* and the Minimum Distance, selection of cut-points was also guided by how the test is intended to be used, and the costs and benefits associated with the test. In situations where the prevalence of a condition may be low, such as FASD, screenings tests are better used to "rule a condition out, but not rule it in" (Streiner, 2003, p. 214), that is to rule out those who definitely do not have the condition. In low prevalence conditions, there is greater confidence that a negative score rules out a condition (Altman & Bland, 1994). Given the focus on developing a screening tool intended to operate on a rule out, rather than rule in principle, sensitivity was prioritized over specificity in the selection of cut-points (Florkowski, 2008).

After cut-points were established, accuracy metrics, as outlined in Table 6.2, were calculated. Accuracy metrics were grouped into 1) those that are independent of the prevalence of the condition, and 2) those that are dependent on the prevalence of the condition. Six accuracy indicators that are independent of the prevalence of a condition are calculated: the AUC, sensitivity, specificity, overall accuracy, the Positive Likelihood Ratio (LR(+)) and Negative Likelihood Ratio (LR(-)). AUC values between 0.5 — 0.7 indicate low accuracy, between 0.7 – 0.9 indicate moderate accuracy, and above 0.9 indicates high accuracy (Streiner & Cairney, 2007). Values of LR (+) greater than 10 and LR (-) less than 0.1 suggest that a test is informative (Fischer, Bachmann, & Jaeschke, 2003). Sensitivity and specificity values of less than 75% or 80% have been suggested as requiring consideration of the applicability of the screening tool (Glover & Albers, 2007), however, decisions around acceptable sensitivity and specificity need to be weighed against the costs and benefits associated with the test.

Table 6.2

Accuracy metric definitions

Accuracy indicator	Definition
Indicators independent of prevalence	
AUC	The probability that a classifier will correctly rank a randomly chosen person with the disease higher than a randomly chosen person without the disease.
Sensitivity	Probability that a test result will be positive when the disease is present (true positive rate).
Specificity	Probability that a test result will be negative when the disease is not present (true negative rate).
Overall accuracy	Overall probability that a patient will be correctly classified
LR (+)	Ratio between the probability of a positive test result given the presence of the disease and the probability of a positive test result given the absence of the disease.
LR (-)	Ratio between the probability of a negative test result given the presence of the disease and the probability of a negative test result given the absence of the disease.
Indicators dependent on prevalence	
PPP	Probability that the disease is present when the test is positive.
NPP	Probability that the disease is not present when the test is negative.
QPPP	Increase in diagnostic value from using this test, taking the prevalence of the condition into account.
QNPP	Increase in diagnostic value from using this test, taking the prevalence of the condition into account.

Note. AUC = Area under the Curve, LR+ = Positive Likelihood Ratio, LR- = Negative Likelihood Ratio, PPP = Positive Predictive Power, NPP = Negative Predictive Power, QPPP= Quality Positive Predictive Power, QNPP = Quality Negative Predictive Power. Sources: (Fischer et al., 2003; Streiner, 2003; Streiner & Cairney, 2007; Usher-Smith, Sharp, & Griffin, 2016).

Four prevalence dependent accuracy indicators were calculated: the Positive Predictive Power (PPP), the Negative Predictive Power (NPP), the Quality Positive Predictive Power (QPPP) and Quality Negative Predictive Power (QNPP). The formula used to calculate the accuracy metrics presented in Table 6.7 and Table 6.8 are presented in Appendix L. As there are no population prevalence estimates for FASD in Australia, multiple prevalence estimates were used based on the estimated prevalence rates found in the general population of school-aged children in the US (May et al., 2018), Canada (Popova, Lange, Poznyak, et al., 2019), and Italy (Aragón et al., 2008; Kodituwakku et al., 2006). For the purposes of this study, accuracy will be

examined at prevalence estimates of 2% and 5%, based on the previously published prevalence estimates and at 1% and 10% to examine performance across a broader spectrum.

The accuracy of the PFST was compared to that of the NST, scored using both scoring algorithms and total score, across the accuracy indicators identified in Table 6.2. Previous research and existing norms were used to establish the cut-points on the NST and SDQ. Individuals were classified as screen positive on the NST if total score ≥ 4 (LaFrance et al., 2014).

Results

Participant Characteristics

Participant characteristics were reported in Chapter 5 (See Table 5.1). A total of 83 responses from caregivers raising children diagnosed with or suspected of having FASD (n = 42) and caregivers from community sample (n = 41) with largely complete data were received. The children from the FASD sample and community sample were similar with respect to age (FASD = 8.07 years (SD = 2.54), community sample = 7.37 years (SD = 2.69), $t(82) = 1.23$, $p = .223$) and gender (FASD = 41.86% female, community sample= 41.46% female, $\chi^2 = .98$, $p = 1.00$).

Reliability

Two indices of internal consistency were calculated based on the 57-item PFST: Cronbach's α and the average inter-item correlation. Cronbach's α was .988 and the average inter-item correlation was .693. The test-retest reliability of the PFST was examined using ICC. For the sub-sample who completed the retest survey (n = 35) the ICC (two-way mixed effects model, with absolute agreement) estimate was .988, 95% CI [.977-.994]. These results indicate that the PFST has excellent test-retest reliability.

Validation

In the following sections, the relationship between the PFST and demographic variables, and the measures included for convergent/divergent validation are examined.

Table 6.3

Observed scale characteristics of the PFST and convergent and divergent validity scales

	N	Missing	Mean	SD	Min	Max	Cronbach's α	Average inter-item correlation (r_s)
PFST	83	0	76.97	57.49	1	166	.988	.693
SDQ Emotional Problems	83	0	3.63	2.71	0	9	.740	.379
SDQ Conduct Problems	83	0	3.48	3.02	0	10	.855	.538
SDQ Hyperactivity	83	0	5.55	3.77	0	10	.926	.729
SDQ Peer Problems	83	0	2.77	2.60	0	10	.802	.438
SDQ Prosocial	83	0	6.73	2.43	2	10	.762	.395
SDQ Total Problem	83	0	15.42	10.14	0	34	.930	.407
NST Total	81	2	4.84	3.44	0	10	.907	.492
CHEXI Working Memory Total	82	1	39.87	19.41	13	65	.987	.836
CHEXI Inhibition Total	82	1	37.17	13.76	12	55	.967	.719
BACC Total	81	2	12.42	10.19	0	35	.928	.387

Note. PFST- Preliminary FASD Screening Tool, SDQ- Strengths and Difficulties Questionnaire, CHEXI- Childhood Executive Functioning Inventory, BAC-C- Brief Assessment Checklist for Children, NST- Neurobehavioural Screening Tool.

Differentiation by known groups results

Scores on the PFST were compared across the age and gender of children. Total scores on the PFST did not differ between male and female participants (U = 674, z = -1.249, p = .212, r = -.139). Further, no relationship was found between the PFST and child age (r = .107, p = .337).

Group differences on the PFST were examined using Mann-Whitney U tests, as shown in Table 6.4. Children with FASD had significantly higher scores than the community sample, indicating greater behaviour concerns on the PFST (FASD mean = 125.22(25.93), Community = 25.37(27.39), U = 41, z = -1.249, p < .001, r = -.820). Further, children with FASD had significantly higher scores than the community sample, indicating greater behaviour concerns on, the SDQ total problems and Hyperactivity, Conduct Problems, Peer Problems and Emotional Problem subscales. The FASD group had significantly lower scores on the SDQ Prosocial scale compared to the community samples indicating greater difficulties with peers in the FASD group. Further, the FASD group also had significantly higher scores on the BAC-C, the NST, and the CHEXI inhibition and working memory indices, as shown in Table 6.4. Effect sizes were moderate to large and ranged from r = .568 on the SDQ prosocial to r = .829 for differences on the NST total score.

Table 6.4

Between groups comparisons of the FASD and community samples of the scales

	FASD			Community				
	Mean	SD	Mdn	Mean	SD	Mdn	U[a]	*Effect size (r)*[b]
PFST Total	125.22	25.93	130.16	25.37	27.39	16	41*	-.820
PFST Retest Total	121.79	23.64	127.15	33.17	41.33	23	24.5*	-.707
SDQ Emotional Problems	5.12	2.24	5.00	2.10	2.27	2	297*	-.568
SDQ Conduct Problems	5.60	2.56	5.00	1.32	1.56	1	125.5*	-.741
SDQ Hyperactivity	8.69	1.89	10.00	2.32	2.10	2	51.5*	-.819
SDQ Peer Problems	4.38	2.31	4.00	1.12	1.69	0	201*	-.670
SDQ Prosocial	5.38	2.21	6.00	8.12	1.79	9	1424.5*	.568
SDQ Total Problem	23.77	5.10	23.00	6.86	5.98	6	59.5*	-.802
CHEXI Working Memory Total	57.00	6.30	58.00	22.73	11.03	18	37*	-.824
CHEXI Inhibition Total	48.41	5.35	49.00	25.93	9.76	24	51*	-.809
BACC Total	20.63	6.94	21.00	4.00	4.46	3	1213*	-.736
NST Total	7.37	2.26	8.00	2.25	2.33	2	41.5*	-.829

Note. PFST- Preliminary FASD Screening Tool, SDQ- Strengths and Difficulties Questionnaire, CHEXI- Childhood Executive Functioning Inventory, BAC-C- Brief Assessment Checklist for Children, NST- Neurobehavioural Screening Tool. Mdn = Median.

[a] Group differences assessed using Mann-Whitney U Tests, due to departures from normality. [b] Effect sizes (*r*) were calculated using the formula $r = Z / \sqrt{N}$ (Field, 2013).

* $p < 0.001$

Convergent validity.

As shown in Table 6.5, results from Spearman rank-order correlations (r_s) indicated that there were strong significant positive correlations between the PFST total score and the SDQ Hyperactivity ($r_s(83) = .873$, $p < .001$) and Conduct Problems ($r_s(83) = .811$, $p < .001$) subscales, and moderate positive correlations between the PFST and the SDQ Emotional Problems ($r_s(83) = .663$, $p < .001$) and SDQ Peer Problems ($r_s(83) = .743$, $p < .001$) subscales. A moderate negative correlation was found between the PFST totals core and SDQ Prosocial subscale ($r_s(83) = -.595$, $p < .001$) scales. Further, the results indicated that there were strong positive correlations between the PFST total score and the CHEXI working memory index ($r_s(82) = .920$, $p < .001$) and the CHEXI inhibition index ($r_s(82) = .905$, $p < .001$). The PFST also showed a high correlation to the NST ($r_s(81) = .845$, $p < .001$). Lastly, the PFST was highly correlated with the BAC-C ($r_s(81) = .876$, $p < .001$). Scatter plots showing the relationship between the PFST and

the scales included for convergent/divergent validation, and identifying the children form the two samples are presented in Appendix M.

Table 6.5

Correlation matrix, Spearman's rank-order correlation (*n* = 81 to 83)

	PFST Total	SDQ Emotional Problems	SDQ Conduct Problems	SDQ Hyperactivity	SDQ Peer Problems	SDQ Prosocial	SDQ Total Problem	CHEXI Working Memory Total [a]	CHEXI Inhibition Total [a]	BAC-C Total[b]	NST Total[b]
PFST Total	–	.663*	.811*	.873*	.743*	-.595*	.909*	.920*	.905*	.876*	.845*
SDQ Emotional Problems		–	.530*	.590*	.526*	-.391*	.763*	.623*	.625*	.658*	.471*
SDQ Conduct Problems			–	.731*	.718*	-.659*	.881*	.763*	.840*	.837*	.897*
SDQ Hyperactivity				–	.631*	-.608*	.866*	.842*	.847*	.820*	.815*
SDQ Peer Problems					–	-.597*	.841*	.721*	.699*	.762*	.710*
SDQ Prosocial						–	-.682*	-.570*	-.615*	-.654*	-.618*
SDQ Total Problem							–	.864*	.880*	.907*	.864*
CHEXI Working Memory Total [a]								–	.882*	.842*	.799*
CHEXI Inhibition Total [a]									–	.878*	.855*
BA-CC Total [b]										–	.841*
NST Total [b]											–

Note. PFST = Preliminary FASD Screening Tool; SDQ = Strengths and Difficulties Questionnaire; CHEXI = Childhood Executive Functioning Inventory; BAC-C = Brief Assessment Checklist for Children; NST = Neurobehavioural Screening Tool.
[a] Analyses of the CHEXI working memory and inhibition scales, *n* = 82, [b] Analyses of the BAC-C and NST, *n* = 81
* = $p < 0.001$,

PFST Screening Accuracy

The accuracy of the PFST to differentiate between children with FASD and the community sample was examined using ROC analysis, followed up by an examination of the PFST's sensitivity, specificity, and predictive power. The accuracy of the NST and SDQ was also examined as comparators. Figure 6.1 shows the ROC curve, for the 57-item PFST. The AUC was .976, 95% CI [.957-1.00], indicating the PFST demonstrated a high level of accuracy in this sample.

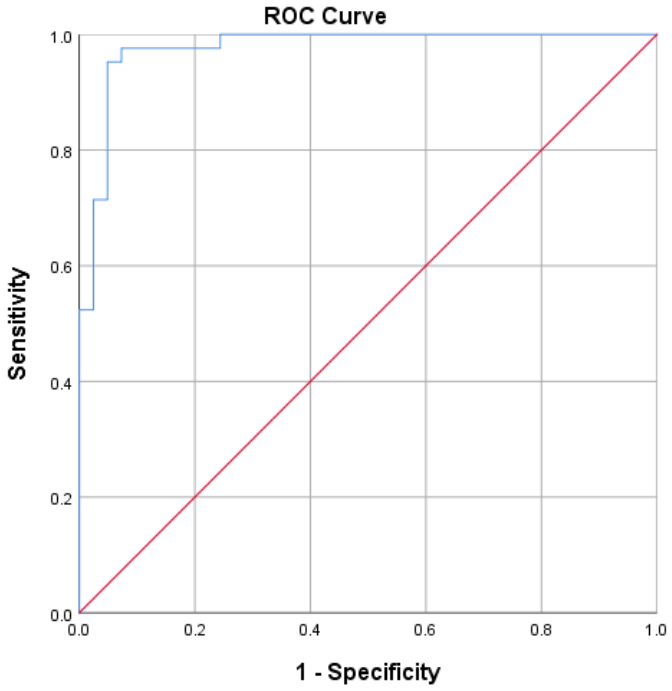


Figure 6.1. ROC Curve for the 57-item PFST.

Youden's *J* and the Minimum Distance were calculated to guide decisions on identifying the optimum cut-point on the PFST to indicate positive or negative screening outcomes. While these indices offer guidance on the selection of cut points, identifying an optimal cut-point requires a subjective judgement about the intended use of the tool and the relative benefits and

costs associated with mis-identified clinical and non-clinical cases. Youden's *J* and the Minimum Distance indices suggested that a cut point of 88 (Youden's *J* = .9036, Minimum Distance = 0.0068) was optimal, yielding a sensitivity = 95.2% and specificity = 95.1%. On inspection, a cut-point of 65 yielded sensitivity = 97.6% and specificity = 92.7%, and demonstrated similar indices *(*Youden's *J* = .9030, Minimum Distance = .077). Comparing the two cut-points, the cut point at 65 had marginally higher sensitivity at the expense of specificity. Given the focus on developing a screening tool intended to operate on a rule out, rather than a rule in principle, sensitivity was prioritized over specificity (Florkowski, 2008; Streiner, 2003) and the cut-point of 65 was selected. The sensitivity, specificity and corresponding Youden's *J* and Minimum distance for each cut-point on the PFST are presented in Appendix N.

Participants were assigned to either "screen positive" if they had a PFST total score greater than or equal to 65 or "screen negative" if PFST total score was less than 65. Participants were assigned to one of four mutually exclusive categories based on their PFST screening outcomes and whether they were diagnosed or suspected of having FASD. Table 6.6 presents the results of the diagnostic classification accuracy of the PFST.

Table 6.6

Classification of results from the validity study of the PFST

		Caregivers reported diagnosed or suspected of having FASD? Yes	No	Row total
PFST screening outcomes	Positive (PFST ≥ 65)	41	3	44
		98%	7%	
	Negative (PFST < 65)	1	38	
		2%	93%	39
Column total		42	41	83

Note. PFST = Preliminary FASD Screening Tool.

Accuracy indicators that are not affected by the prevalence of a condition are presented in Table 6.7. As shown in Table 6.7, the PFST shows a high level of sensitivity (97.6%) and specificity (92.7%). Based on the cut point of 65, the PFST had an overall classification accuracy rate of 95%. The likelihood ratio (LR+ = 13.34) suggests that a positive score on the PFST is approximately 13 times more likely for those who have FASD than those who do not. The negative likelihood ratio (LR- = 0.026) suggests a negative result is unlikely to have come from an individual who does not have FASD.

Table 6.7

Accuracy indicators that are not affected by prevalence

Indicator	Value	95% CI
AUC	.976	[.957,1.00]
Sensitivity	97.6%	[87.4%, 99.9%]
Specificity	92.7%	[80.1%, 98.5%]
Overall accuracy	95.18%	[88.21%, 98.67%]
LR+	13.34	[4.48, 39.70]
LR-	0.026	[0.00, 0.18]

Note. AUC = Area Under the Curve, LR+ = Positive Likelihood Ratio, LR- = Negative Likelihood Ration, CI = Confidence Interval.

Several indicators of accuracy are influenced by the prevalence of the condition being examined. In this study, the apparent prevalence rate was 51% (42 participants with FASD, and 41 community participants). As noted above, if PPP and NPP were calculated using the prevalence of the samples in this study, (PPP = .932 and NPP = .974), which would suggest that the PFST is accurate in 93% of screen positive cases. As no accurate population prevalence rates for FASD are available for Australia, these accuracy metrics dependent on prevalence rates were calculated using estimates of 1%, 2 %, 5%, 10% based on studies conducted in the United States and Canada (May et al., 2018; Popova, Lange, Poznyak, et al., 2019) and the prevalence in the current sample (51%). Table 6.8 displays the PPP, NPP, and QPPP and QNPP for the PFST. As shown in Table 6.8, the PPP of the PFST is .214 at an estimated 2% prevalence, indicating that,

in a population with 2% prevalence, 21.4% of positive screens would come from someone with behavioural difficulties indicative of FASD, and the other 78.6% would be false positives. Similarly, the NPP indicates that, at a 2% prevalence rate, 99.9% of negative cases would be true negatives. As the prevalence of a disorder increases, the PPP also increases, and at an estimated prevalence rate of 10%, the predictive power of the PFST was .597, indicating that 59.7%, of positive screens would indicate would be true positives.

Table 6.8

Prevalence-dependent accuracy metrics for the PFST across a range of estimated prevalence rates and the present study

	Estimated Prevalence Rates				
Accuracy Metrics	1%	2%	5%	10%	Present study (Prevalence = 51%)
PPP	.119	.214	.413	.597	.932
NPP	.999	.999	.999	.997	.974
QPPP	.110	.198	.382	.552	.862
QNPP	.974	.974	.973	.972	.949

Note. PPP. = Positive Predictive Power, NPP = Negative Predictive Power, QPPP = Quality Positive Predictive Power, QNPP = Quality Negative Predictive Power.

Comparisons of screening accuracy with the NST

Table 6.9 shows the classification contingency tables for the NST-A (FASD from typically developing peers) and NST-B (FASD without ADHD symptoms from typically developing peers), NST total scores, and the SDQ. The classification outcomes of the PFST are repeated for reference. Results are presented in Table 6.9 for the NST-B algorithm but no further calculations were conducted on the NST-B accuracy as no cases were screen positive.

Table 6.9

Classification outcomes of the NST, scored using the algorithm and total scores

		Diagnosed or suspected of having FASD	
	Screening outcome	Yes	No
PFST	Positive	41	3
(*n*=83)	Negative	1	38
NST A	Positive	15	1
(n=81)	Negative	26	39
NST-B[a]	Positive	0	0
(*n*=81)	Negative	41	40
NST Total[b]	Positive	40	9
(n=81)	Negative	1	31

Note. PFST = Preliminary FASD Screening Tool, NST A = NST, scored using the author supplied algorithm, NST B = Scored using the author supplied algorithm for children with FASD who do not display ADHD symptoms.
[a] The NST-B identified children who display behaviour suggestive of FASD without ADHD symptomatology
[b] Cases classified as screening positive on NST total if scoring ≥ 4 (LaFrance et al., 2014)

Table 6.10 shows the accuracy of the PFST, and the NST as scored using the algorithm for the NST-A and total score. As shown, the PFST demonstrated higher overall accuracy (95.2%, 95% CI [88.2% – 98.7%]) rates than the NST as scored using both the algorithm (66.7% 95% CI [53.3% – 76.8%]) and the total score (87.7%, 95% CI [78.5% – 94.0%]).

Table 6.10

Accuracy metrics, not affected by prevalence across measures[a]

	PFST		NST A		NST Total	
	Estimate	95% CI	Estimate	95% CI	Estimate	95% CI
AUC	.976		[b]		.925	
Sensitivity	97.6%	[87.4% - 99.9%]	36.6%	[22.1% - 53.1%]	97.6%	[87.1% - 99.9%]
Specificity	92.7%	[80.1% - 98.5%]	97.5%	[86.4% - 99.9%]	77.5%	[61.2% - 89.2%]
Overall Accuracy	95.2%	[88.2% - 98.7%]	66.7%	[53.3% - 76.8%]	87.7%	[78.5% - 94.0%]
LR +	13.34	[4.48 - 39.70]	14.63	[2.03 - 105.66]	4.34	[2.42 - 7.72]
LR -	0.026	[0.00 - 0.18]	0.650	[0.51 - 0.82]	0.031	[0.00 - 0.22]

Note. AUC = Area Under the Curve, LR+ = Positive Likelihood Ratio, LR- = Negative Likelihood Ratio, PFST = Preliminary FASD Screening Tool, NST A = NST, scored using the author supplied algorithm.
[a] Accuracy metrics could not be calculated for the NST-B as no suspected cases of FASD without ADHD symptomatology were identified using the NST in this sample. [b] AUC not calculated, as the algorithm outcome is dichotomous.

The accuracy of the PFST and the NST were compared across the estimated prevalence rates (1%, 2%, 5% and 10%). Table 6.11 shows the accuracy metrics that are dependent on prevalence at each prevalence estimate for the measures. As shown in Table 6.11, the PPP of all tests is relatively low at low prevalence, indicating a low probability that a positive score has FASD. While the NST-A shows higher PPP and QPPP across the estimated prevalence rates, the NPP, and quality PPP is lower, suggesting that there is a low probability that FASD is not present when the test is negative (i.e., less likely to identify individuals who do have FASD), and this is supported by the low sensitivity noted in Table 6.10. As expected, the PPP and QPPP increase for all measures as the estimated prevalence increases.

Table 6.11

Prevalence dependent accuracy metrics [a]

	PPP	QPPP	NPP	QNPP
Estimated prevalence = 1%				
PFST	.119	.110	.999	.974
NST-A	.129	.120	.993	.347
NST Total	.042	.032	.999	.968
Estimated prevalence = 2%				
PFST	.214	.198	.999	.974
NST-A	.230	.214	.987	.345
NST Total	.081	.079	.998	.910
Estimated prevalence = 5%				
PFST	.413	.382	.999	.973
NST-A	.435	.405	.967	.338
NST Total	.186	.143	.998	.967
Estimated prevalence = 10%				
PFST	.597	.552	.997	.972
NST-A	.619	.577	.933	.326
NST Total	.325	.225	.997	.965

Note. PFST = Preliminary FASD Screening Tool, NST-A = NST Scored by algorithm, NST = Neurobehavioural Screening Tool, PPP. = Positive Predictive Power, NPP = Negative Predictive Power, QPPP = Quality Positive Predictive Power, QNPP = Quality Negative Predictive Power.
a = Accuracy metrics could not be calculated for the NST-B as no suspected cases of FASD without ADHD symptomatology were identified using the NST in this sample.

Discussion

This chapter sought to examine the reliability, validity, and screening accuracy of the PFST. The PFST demonstrated an acceptable level of internal consistency, as assessed using Cronbach's alpha and the average item-total correlation. While Cronbach's alpha was high (α = .988), this may be reflective of the length of the tool, as of sound internal consistency (Streiner et al., 2015) as Cronbach's alpha can be inflated when a scale contains a large number of items (Streiner et al., 2015). As a result, this result should be interpreted with caution. The average inter-item correlation (.693) was somewhat higher than the range recommended by Clark and Watson (1995). Taken together, the PFST showed an acceptable level of internal consistency. A conservative approach was taken to item selection for preliminary reliability and validity testing, and potential future research directions to further refine the content of the PFST are discussed.

As expected, scores on the PFST differed between children previously diagnosed with or suspected of having FASD and a community sample of children. Total scores on the PFST showed high, positive correlations with measures of working memory, inhibition, hyperactivity, conduct problems, peer relationships and emotional problems. The high correlations demonstrated with these constructs suggests that the PFST is measuring a range of behavioural and cognitive domains relevant to the functioning of children with FASD. Children with FASD demonstrate impairments in a broad range of cognitive and behavioural domains (see Chapter 2). The scales used for convergent/divergent validation of the PFST in this preliminary study measure several different, albeit related concepts.

The PFST appears to capture multiple dimensions of cognitive and behavioural functioning, as evidenced by the high correlations with the measures included for convergent/divergent validation. Children with FASD demonstrate deficits in broad range of cognitive and behavioural domains (See Chapter 2, (Mattson et al., 2019)). The PFST was

developed to reflect this broad cognitive and behavioural profile. In contrast to other screening tools that have been developed for children with FASD (e.g., the Personal Behaviour Checklist/Fetal Alcohol Behaviour Scale (Streissguth et al., 1998), and the NST (Nash et al., 2006), there has been limited investigation of the relationships between the screening tools and related constructs.

The high correlation between the PFST and the SDQ, CHEXI, and BAC-C may represent the inclusion of similar concepts across measures. Scores on the PFST were unrelated to age and did not differ by gender, suggesting the tool may be used for both males and females and across the age range of interest. When examining the accuracy of the screening tool, the PFST demonstrated a high level of sensitivity (97.6%) and specificity (92.7%) in this sample. Accuracy estimates were calculated at various prevalence rates, given that the positive and negative predictive power is dependent on prevalence, and the approximately 50% prevalence in this sample is not representative of the wider population. The PFST demonstrated strong NPP across prevalence estimates, but low PPP suggesting large numbers of false positives when the prevalence is low in the population and highlighting the potential need to follow up positive scores on the PPP with a second test with higher specificity.

As a screening tool, the PFST cannot be used to diagnose children with FASD, but rather can be used to identify children with a cognitive-behavioural profile consistent with FASD and in need further assessment. In developing the PFST, two potential cut-points for screening positive or negative were identified. The cut-point of screening positive if the PFST score ≥ 65 was selected as it had a higher sensitivity (at the expense of a lower specificity) compared to screening positive if the PFST score was ≥ 88. As FASD is a low prevalence condition, screening tools should adopt a “rule out” rather than a “rule in” approach, and aim for a higher sensitivity to minimise false negatives (Streiner, 2003). In low prevalence

conditions, there is greater confidence that a negative score rules out a condition (Altman & Bland, 1994).

The PFST demonstrated a high level of accuracy in differentiating between children between the ages of four and 12 based on their cognitive and behavioural profile. This highlights the potential utility of the PFST as a screening tool to identify children who display cognitive and behavioural difficulties consistent with children who have FASD. There are, however, several explanatory mechanisms that may be contributing to the differences noted across studies including comorbid exposure to trauma, and the representativeness of the sample.

Potential explanatory mechanisms

Comorbid exposure to trauma

As reported in the previous chapter, the majority of children with FASD in this study were in the care of a caregiver who was not a biological parent (92.86%) compared to 9.76% of children in the community sample. This is consistent with previous literature involving children with FASD reporting high proportions of children no longer in the care of their birth parents. Studies identified in the literature review (Chapter 2) report between 55% to 100% of children with FASD live with caregivers other than their biological parents. Furthermore, epidemiological studies suggest FASD may be over-represented amongst children who are removed from their birth parents care (Fuchs et al., 2010; Popova, Lange, Poznyak, et al., 2019). In the present study, children with FASD were reported to experience high levels of attachment- and trauma-related mental health difficulties commonly observed among children in foster, kinship, residential and adoptive care, as measured by the BAC-C, whereas the community sample displayed low levels of mental health symptomatology. It is not known whether children in the FASD sample who were not living with their biological parents were the subject of child protection or family court orders or were living in informal arrangements

and whether these children had experienced abuse or neglect in childhood, in addition to PAE. Such experiences may significantly impact on the cognitive and behavioural development of children (D'Andrea et al., 2012; McLaughlin et al., 2017) and may contribute to between group differences reported on the PFST, the SDQ and subscales, and the CHEXI. Questions directly enquiring about trauma exposure were not, however, included in the questionnaire. Several participants did report that their children with FASD had a comorbid PTSD diagnosis.

Sample Representativeness

As noted in Chapter 5, the FASD group and the community sample differed significantly with respect to caregiver education. Approximately 97% of the community sample reported undergraduate or post-graduate education compared to the FASD group where approximately 43% reported undergraduate or post-graduate education, reflecting potential sampling bias in the community sample. These differences may be contributing to large difference noted between the groups across the measures included in the initial reliability and validity study and be a further contributing factor to the high levels of accuracy between the groups. Parental education is a significant protective factor for child development (Duncan & Magnuson, 2012; Walker et al., 2011), and the impact of the sample bias will be further discussed in Chapter 7.

The prevalence of the condition being examined in studies of screening or diagnostic accuracy can bias the results. The prevalence of FASD has been estimated at between 2% and 5% of the general population (May et al., 2018; Popova, Lange, Poznyak, et al., 2019). The prevalence of the present sample (51%) may not reflect the prevalence in the wide community, as a result the accuracy rates of the PFST may be inflated (Kohn et al., 2013). Further, it is noted that screening tools perform best when the prevalence rate is approximately 50% (Streiner, 2003). While the PPP and NPP were examined at estimated

prevalence rates, future studies in a sample more representative of the population prevalence of FASD will be required to validate the accuracy of the PFST.

Comparison with the NST

The accuracy metrics of the PFST were compared to the NST. The PFST had higher sensitivity, classification accuracy, and NPP across estimated prevalence rates than the NST, particularly when scored using the author-supplied algorithm. The NST-B algorithm, which is intended to identify children who display behaviour suggestive of FASD without ADHD symptomatology, did not identify any participants as "screen positive" in this sample. This may reflect a high level of symptomatology comorbid with ADHD among the FASD group. The scatterplots of the SDQ (Appendix M) support high levels of hyperactivity among the FASD group.

The accuracy metrics of the NST when scored using the total score were higher than when scored using the algorithm. Compared to the algorithms which require a specific pattern of scoring across items (e.g., at least 6 yes responses to NST items 1-7, and at least 3 yes responses to items 1, 8-10 for screen positive) the total score represents a more liberal cut-point as the total score makes no assumptions about the pattern of scoring, instead treating items equally.

Limitations

A limitation of this study is the lack of a "gold standard" FASD diagnostic tool against which to compare the accuracy of a potential screening tool for children previously diagnosed with FASD. Limitations of the gold standard in developing screening and diagnostic tools are not unique to FASD (Streiner, 2003). With regards to FASD, the gold standard to assess against in Australia would be limited to the multi-disciplinary assessment as outlined in the Australian FASD Diagnostic guidelines (Bower & Elliott, 2016). However, access to such services is limited due to the lack of clinicians with the expertise, and capacity

in diagnostic clinics. Furthermore, given the relatively recent publication of these guidelines, some children's diagnoses may have preceded the publication of these guidelines meaning they may have been diagnosed under varying guidelines, which include different definitions and cut-offs for determine cognitive and behavioural impairment (Coles et al., 2016). In the present study, caregivers reported a range of diagnostic processes including multi-disciplinary FASD assessments, single clinicians making a FASD diagnosis, and for some, they were still awaiting assessment aspects and diagnosis confirmation. For the purposes of this study, cases were defined based on confirmed or pending diagnosis. There is a possibility that on completing the diagnosis assessment, some children included in the FASD group in the current study would be found not to have FASD. Further, as children varied by the length of time since they were diagnosed and may have had access to varying levels of support following diagnosis, this could serve to reduce cognitive and behavioural concerns experienced by families by having support for these concerns. Living in a stable home environment has been noted to improve the long-term outcomes for individuals with FASD (Streissguth et al., 2004). Finally, the fixed order of presentation made some tools less likely to be completed, and thus could introduce systematic bias.

Future Research Directions

Future research using the PFST could seek to extend these promising preliminary findings by examining the performance of the PFST in larger samples and using analytical methods such as factor analysis to further refine the content of the PFST. Future research should seek to reduce the sampling bias in community comparison groups. Testing the PFST in larger, more diverse samples including children with a range of cognitive and behavioural concerns that is more representative of the settings where the tool will ultimately be used will be vital in determining the PFST accuracy. Previous research has found good sensitivity and specificity when comparing children with FASD to typically developing peers, but lower

specificity when comparing children with FASD to other neurodevelopmental disabilities (Lange et al., 2019). Examining the sensitivity and specificity of the PFST in a more diverse population including children with other neurodevelopmental disabilities and exposure to trauma but without FASD will provide important information about how the PFST may be applied in practice in specialist and universal services. Future research linking with Active Case Ascertainment studies to provide a confirmed diagnosis will be important for further PFST development.

The underlying prevalence and its impact on accuracy metrics needs to be considered for future research. Accuracy of screening tools is best when the prevalence of a condition is approximately 50% (Streiner, 2003), as it is in this sample. However, this does not represent the general population, and consequently the PFST should be examined further in a representative sample setting where the tool may be used to ensure test accuracy is not over-estimated (Usher-Smith et al., 2016). As noted, the PPP and NPP are dependent on the prevalence rate of the condition being examined, and screening tools appear to perform best when the prevalence is approximately 50% in the population being tested. In low prevalence settings (e.g. universal settings such as preschools or schools), a greater number of false positives may be identified. In more specialised settings where the prevalence may be higher (e.g., behavioural clinics or among children in out of home care), the false prevalence rate may be lower. However, with such high convergence with other tools used in these settings (e.g., SDQ, CHEXI, BAC-C) further testing of the PFST in representative and specialised settings is needed.

As the current version of the PFST contains 57 items this may limit is applicability in certain settings, as well as confound the interpretation of the internal consistency statistics. In addition to examining the performance of the tool in broader settings, future research could seek to examine the factor structure of the tool. Factor analysis was not undertaken in the

present study due to sample size considerations. Factor analysis may also be used to reduce the length of the PFST by only retaining those items that adequately load onto factors. Lastly, future studies could seek to further examine the validity of the PFST by examining the convergent/divergent validity against a broader set of constructs, including consideration seeking different observers (e.g., teachers), or direct testing of related constructs with children using the multi-trait, multi-method approach (Streiner et al., 2015).

Conclusions

The results of this pilot study suggest that the PFST shows promise as a tool for identifying children who may be at risk of having FASD. Measure development is a lengthy and on-going process. Further refinement and development of the PFST is required before implementation or scale up. The PFST demonstrated a high level of accuracy in differentiating between children with FASD and the community sample. The PFST demonstrated an acceptable level of reliability and showed high correlations in the expected direction with measures of related concepts including executive functioning and behaviour concerns. There were, however, some limitations noted, particularly the recruitment bias present in the community sample, and the high level of mental health concerns potentially reflecting exposure to trauma. Further research using the PFST is needed to strengthen and confirm psychometric properties before the PFST can be implemented and used. Chapter 7 will discuss the overall findings and further examine the overlap of FASD and trauma as noted in Chapter 5 and 6. The strengths and limitations of the PFST will be discussed and directions for future research identified.

Chapter 7 Discussion

The aim of the research reported in this thesis was to develop a screening tool for children between the ages of 4 and 12 years old to identify those who display cognitive and behavioural impairments consistent with FASD. The aim was to develop a tool that would be quick and easy to use, and cover the breadth of cognitive and behavioural impairments experienced by children with FASD. A sequential measure development process (Figure 7.1) was followed to develop the Preliminary FASD Screening Tool (PFST). This process involved drawing on the existing literature and conducting interviews with caregivers and clinicians to inform the content of the screening tool, before further refining the content of the tool through quantitative item analysis. The final stage of this process involved conducting a preliminary study of the reliability and validity of the PFST. The following chapter summarises the findings of each chapter, and discusses the implications for screening for FASD, the strengths and limitations of the research, and avenues for future research.

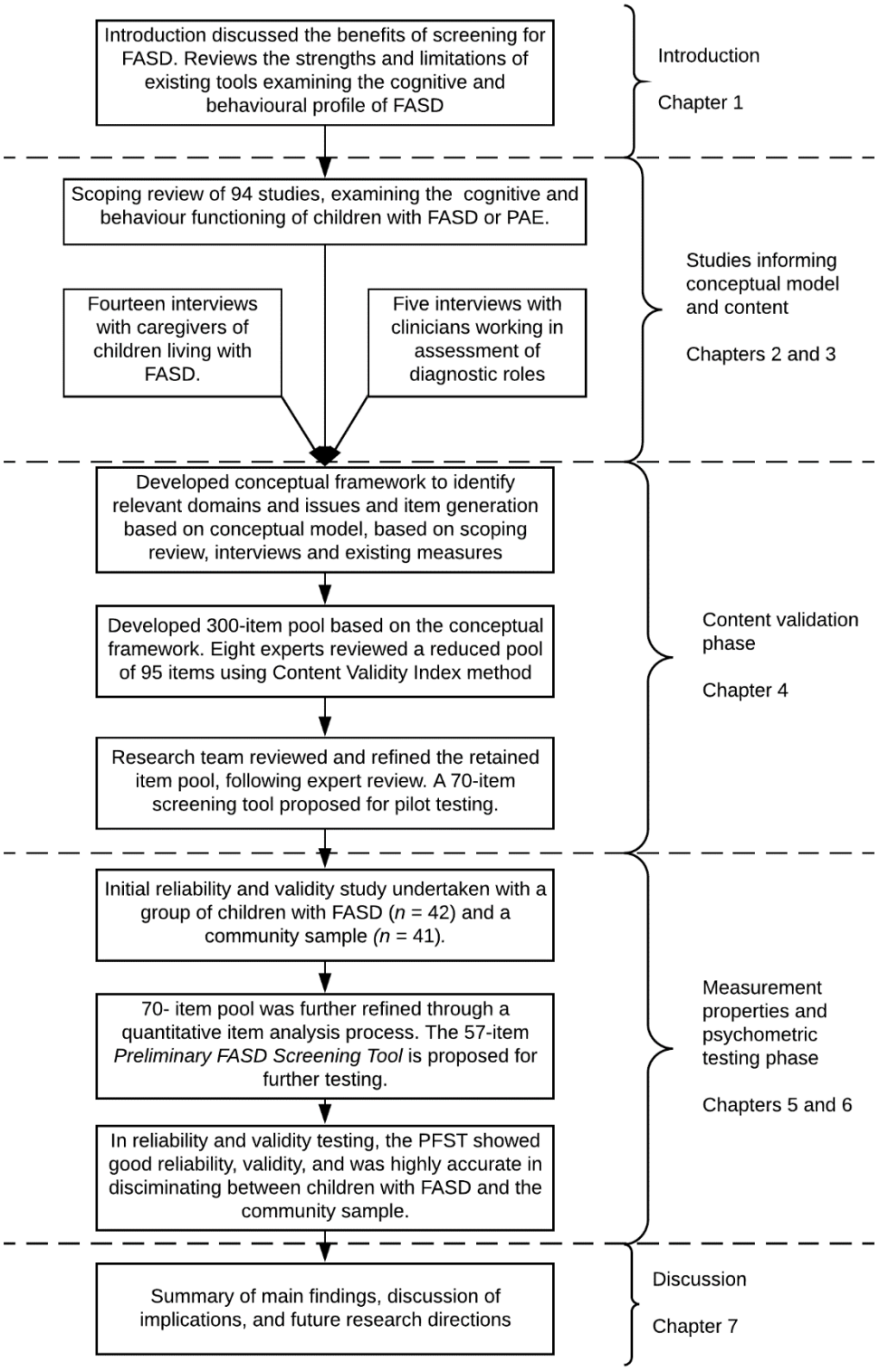


Figure 7.1: Overview of thesis methodology

Summary of Main Findings

Chapter 2: Scoping Review

A scoping review of the published literature was undertaken. Ninety-four studies examining the cognitive and behavioural impairments associated with FASD and PAE among children between the ages of three and 13 were identified. Studies were split into four methodological categories: (a) studies comparing outcomes for children diagnosed with FASD (*n* = 50), (b) studies of children with PAE compared to non-exposed peers (*n* = 9), (c) longitudinal studies of the outcomes of PAE among children split into different levels of PAE (*n* = 22), and (d) longitudinal studies examining the relationship between alcohol exposure as a continuous variable (e.g., average alcohol consumption per day) and subsequent outcomes (*n* = 13). Across studies, children with FASD and children heavily exposed to alcohol prenatally were found to display impairments in the domains of academic achievement, adaptive behaviour, attention, executive functioning, language, social skills, sleep, and sensory processing. Impairments in these domains were often reported from a young age. Among the studies examining outcomes among children diagnosed with FASD, or comparing children with PAE to non-exposed peers, large proportions (17.5%-100% in the 31 studies to report this information) of children with FASD, or children with heavy PAE were reported to no longer be in the care of their birth parents. Lower proportions of children living with carers other than biological parents were noted among the South African studies. While consistent patterns of impairments were found across many studies, studies of children with FASD frequently included high proportions of children who are no longer in the care of their birth parents, and the potential overlap with experiences of abuse and neglect may impact upon cognitive and behavioural functioning of children with FASD and PAE.

Chapter 3: Interview Study

Chapter three described a qualitative study examining the cognitive and behavioural functioning of children with FASD from the perspectives of their caregivers and clinicians who work in assessment, diagnostic, or therapeutic roles. Fourteen caregivers and five clinicians (two paediatricians, one clinical neuropsychologist, one speech pathologist, and one occupational therapist) participated in the study. Clinicians proved difficult to recruit, owing in part to the small numbers of clinicians with expertise in the diagnosis and assessment of FASD in Australia. Caregiver descriptions of difficulties experienced by preschool and primary school aged children with FASD were coded into three over-arching themes, and several major subthemes: self-regulation (behavioural regulation, emotional regulation, and attention regulation), cognitive ability (memory and academic ability), and adaptive functioning (communication and language, social skills, motor skills, sleep concerns). Caregivers also described their children as acting young for their age and demonstrating inconsistency in behaviour across time. Caregivers identified concerns over behavioural and emotional regulation as early warning signs that prompted them to seek assessment or support. Clinicians commonly identified difficulties with behavioural regulation, cognitive ability (particularly difficulties learning from mistakes) among the children with FASD with whom they worked. Reflecting their professional expertise, the occupational therapist and speech pathologist spoke extensively about difficulties in motor skills and language, respectively. Clinicians and caregivers spoke to a number of consistent difficulties including difficulties with hyperactivity, impulsivity, not learning from mistakes. The situational specificity of some behaviours (e.g., tantrums and meltdowns, difficulties with sleep) may account for why these behaviours were not discussed in clinicians' interviews.

Chapter 4: Item Development and Content Validation

The fourth chapter described the process of developing the item pool and refining this through review by an expert panel. A hypothesised framework of the cognitive and behavioural domains most salient to include in a FASD screening tool was developed from the findings of the scoping review and the interviews with caregivers and clinicians. This framework informed the development of the initial item pool. A conservative approach was taken when refining items in this chapter, as items could be further refined or excluded in subsequent testing. An initial pool of 300 items was generated based the domains of interest. After an initial review by the research team, a pool of 95 items measuring the domains included in the framework were reviewed by a panel of experts, including six caregivers raising children with FASD and two clinicians with expertise in the assessment and diagnosis of FASD. The experts rated the 95 items on their relevance for inclusion in a screening tool for children who may have FASD and provided comments on anything that may have been missed. The Content Validity Index (CVI) was calculated, and items with low CVI were reviewed for potential removal from the tool. Sixty-eight items were retained, showing good CVI at the item and scale level. Further refinements based on the expert panel review were undertaken through a review by the research team, and a 70-item pool was retained for further testing. The 70 items reflected a range of cognitive and behavioural domains including adaptive behaviour (communication, motor skills, social skills, sleep, and young for age), cognitive functioning (academic abilities, general cognitive functioning, and learning and memory), and self-regulation (attention regulation, behavioural regulation, and emotional regulation).

Chapter 5: Quantitative Item Analysis

The fifth chapter described the methodology and results of the preliminary reliability and validity testing of the quantitative item analysis used to further refine the items included

in the PFST. Data gathered from a preliminary study of 83 participants (n = 42 caregivers raising a child with FASD, and n = 41 caregivers raising children from a community sample) was used to further refine the item pool developed in Chapter 4. The quantitative item analysis process examined the item discrimination, item-total correlation, and average inter-item correlations. Of the 70 items proposed for inclusion in the PFST, 57 items were retained based on the results of the quantitative item analysis process. The retained items represented the three overarching themes identified in the interviews: adaptive functioning (e.g., social skills, language, *n* = 20 items), cognitive functioning (e.g., academic performance, memory, *n* = 16 items) and self-regulation (e.g., behavioural regulation, emotional regulation, and attention regulation; *n* = 21 items). Items relating to behavioural regulation such as hyperactivity and, impulsivity; and those relating to cognitive functioning, particularly around working memory, attention, and understanding of consequences were retained, and showed good item discrimination between the children with FASD and the community sample in the pilot study.

Chapter 6: Preliminary Reliability and Validity

Chapter six described the results of the overall preliminary reliability and validity study. The sample who participated in the quantitative item analysis (Chapter 5) were retained for the preliminary reliability and validity study. The 57-item PFST demonstrated good test-retest reliability (ICC = .988, 95% CI: .977 - .994) and acceptable internal consistency (Cronbach's α = .988, average inter-item correlation = .693). Mean scores on the PFST did not differ by gender. Further, no relationship was found between PFST scores and child age.

The PFST demonstrated a high level of convergent validity with a range of tools that were selected to reflect the domains of interest on the PFST. These included working memory and inhibition as measured by the Childhood Executive Functioning Inventor (CHEXI),

hyperactivity as measured by the SDQ, more general measures of emotional regulation as measured SDQ Emotional Problems and Peers Problems subscales, behavioural functioning as measured by the SDQ Conduct Problems, FASD-specific measures as measured by the NST, and measures of trauma symptoms as measured by the Brief Assessment Checklist-Children (BAC-C). The PFST demonstrated high correlations with measures of executive functioning (CHEXI WM r_s = .920 and Inhibition r_s = .905) and behavioural functioning (SDQ Hyperactivity r_s = .873 and Conduct Problems r_s = .811). Moderate correlations were reported between the PFST and measures of emotional functioning (SDQ Emotional Problems r_s = .663 and Peer Problems r_s =.743). Further, the PFST was negatively related to the SDQ Prosocial subscale (r_s = -.595) as expected. The PFST demonstrated a high level of accuracy in differentiating between children diagnosed with or suspected of having FASD and children in the community sample (sensitivity = 97.6%, specificity = 92.7%, LR+ 13.34, LR - 0.026). Compared to the NST, as scored using the author-supplied algorithms, the PFST demonstrated higher overall accuracy (PFST = 95.2%, NST A = 66.7%) and sensitivity (PFST = 97.6%, NST = 36.6%), and the specificity of both tools was similar (PFST = 92.7%, NST = 97.5%). Compared to the NST screening outcomes defined by total score (LaFrance et al., 2014), the PFST demonstrated higher overall accuracy (PFST = 95.2%, NST A = 87.7%) and specificity (PFST = 92.7%, NST = 77.5%), and the sensitivity of the tools was the same (PFST = 97.6%, NST = 97.6%).

Summary of Overall Findings

This thesis set out to develop a new screening tool for FASD, following measure development guidelines, that would be quick, easy to administer and which covered the breadth of cognitive and behavioural impairments experienced by children with FASD. This study followed an established process of measure development (Boateng et al., 2018; Streiner et al., 2015) to ensure that the content of the PFST reflected the breadth of cognitive and

behavioural functioning associated with FASD. The overall findings were that the PFST demonstrated a high level of sensitivity and specificity, and excellent test-retest reliability. The findings of this series of studies have also been examined in the context of sampling and other limitations, and further research is suggested to examine the validity of the PFST including factor analysis. What follows is a description of the overall strengths of the study, consideration of potential limitations, clinical implications and suggestions for future research.

Strengths of the Study

While existing screening tools for identifying children who display the cognitive and behavioural impairments consistent with FASD (e.g., the NST (Nash et al., 2011; Nash et al., 2006) and PBCL-36/FABS (Streissguth et al., 1998)) have been proposed, a number of limitations of these tools were identified (See Table 1.1). The NST was developed by drawing content only from the CBCL, and as a result, reflects a narrow view of the difficulties experienced by children with FASD. Further, no studies appear to have been conducted examining the reliability and validity of the NST beyond those that have examined the accuracy of the NST. The PBCL-36, while developed incorporating extensive clinician and caregiver experiences, appears to have limited subsequent studies examining the reliability and validity, and where the PBCL-36 has been used, the accuracy in identifying children who may have FASD has not been examined.

The development process followed in creating the PFST addressed a number of the limitations associated with these measures. The development process of the PFST drew on established measure development procedures. The content of the PFST was developed through an extensive literature review, interviews with caregivers raising children with FASD, and clinicians with expertise in diagnostic and assessment roles. By including caregivers' and clinicians' perspectives and focusing on impairments that may emerge in a

child's preschool and primary school years, the PFST identified a broad range of impairments associated with FASD that can assist early detection and the potential for early intervention. The cognitive and behavioural difficulties highlighted in the interviews were consistent with previous qualitative research with caregivers examining the cognitive and behavioural functioning of children, adolescents, and young adults with FASD (Gardner, 2000; Salmon, 2008; Sanders & Buck, 2010; Whitehurst, 2012). An extensive item pool covering the over-arching domains of self-regulation, cognitive functioning, and adaptive functioning was developed. Further, a number of items in the tool reflect self-regulatory difficulties and there is substantial evidence that self-regulation difficulties underpin the behavioural presentation of FASD (Reid & Petrenko, 2018) and a number of interventions for children with FASD focus on improving self-regulation (Reid et al., 2015). Feedback from an expert panel comprised of caregivers and clinicians suggested that the PFST had good content validity, and the item pool was refined to 70-items for inclusion in a preliminary reliability and validity study.

The content of the PFST underwent further refinement in a preliminary reliability and validity study, using a quantitative item analysis process, to ensure that the items retained are functional and internally consistent (Boateng et al., 2018). The validity of the PFST is also supported by the results of qualitative item analysis, in particular, the strong item discrimination as judged against the external criterion of whether or not the individual had, or was suspected of having, a FASD diagnosis (Urbina, 2004). The 57-item PFST showed excellent test-retest reliability and adequate internal consistency, noting however, there may be some redundancy among the retained items. Validity was examined by comparing the scores on the PFST across known groups (e.g., children with or suspected of having FASD, compared to the community sample), by examining the relationships with a wide range of related constructs (e.g., executive functioning, behavioural and emotional functioning) in a

convergent validity method, and by examining the performance of the tool in differentiating between children with FASD and a community sample of typically developing peers. Taken together, these results suggest that the PFST demonstrates adequate content validity (i.e., covers the breadth of cognitive and behavioural difficulties experienced by FASD), and shows promising preliminary reliability and validity. The PFST demonstrated high sensitivity and specificity in differentiating the children with FASD from the community sample.

The PFST appeared to have greater accuracy than the NST, as demonstrated by the higher overall accuracy, higher sensitivity and specificity. Further, the positive and negative likelihood odds ratios lie within the recommended range for informative tests (Fischer et al., 2003). While scoring the NST using the algorithm resulted in marginally better positive predictive power that the PFST, the sensitivity and negative predictive power were considerably lower, suggesting that when the NST is used, there is a low probability that an individual did not have FASD when then test is negative and that a large number of false negatives would be generated. Given that a screening tool for low prevalence conditions is best utilised to operate on a "rule in" principle (Streiner, 2003), a high level of false negatives is of concern to the utility of the NST in practice. The NST and PFST scored similarly when the NST was scored using the total score with regards to the negative predictive power, however the PFST showed greater positive predictive power across the estimated prevalence rates, suggesting that the PFST may identify fewer false positives at lower prevalence rates, which is an important outcome for the feasibility of a screening tool. A lower positive predictive value is associated with a greater number of false positives, suggesting that a tool is over-identifying individuals as at risk (Glover & Albers, 2007).

Of note, the NST-B pathway, which is intended to capture children with FASD who do not display ADHD symptoms, identified no cases in the present sample as screen positive. This may reflect the high proportions of children in the FASD sample who had high levels of

hyperactivity symptoms as per the SDQ Hyperactivity scale (See Appendix M). The PFST incorporates a broader range of behaviour and demonstrated good content validity, as rated by experts, and good psychometric properties. Although the PFST is considerably longer than the NST, the additional time taken must be balanced against the poorer performance, especially if scored using the algorithm that is advocated. Further research, as outlined below, may serve to further refine the content, and reduce the length of the PFST, thereby overcoming some of the participant burden.

While the PFST was able to discriminate with high levels of accuracy between two samples – one of children with FASD (confirmed or provisional diagnosis) and a community sample, there was an element of sampling bias noted in the preliminary reliability and validity study. Consistent with other studies that have used similar recruitment methods (Price, 2019; Reid & Moritz, 2019), the FASD group included children who were living with kin and foster carers and who had had experiences of childhood trauma. Further, the community sample included caregivers with high levels of education attainment in comparison with the FASD sample and the general population. The following sections will examine the implications of this for interpreting the findings of the current study in the broader context of understanding the cognitive and behavioural profile of children with FASD, and for future research.

The Cognitive and Behavioural Profile of FASD

As identified throughout this thesis, children with FASD experience deficits in a range of cognitive and behavioural domains. Research has sought to identify a distinct neurodevelopmental profile associated with FASD, defined “as the outward expression (behavioural and developmental) of the central nervous system damage associated with prenatal alcohol exposure” (Lange et al., 2017, p. 2). Currently, the Australian diagnostic assessment of FASD considers ten domains of cognitive and behavioural functioning (i.e.,

brain structure/neurology, motor skills, cognition, language, academic achievement, memory, attention, executive functioning, affect regulation, and adaptive behaviour, social skills or social communication) (Bower & Elliott, 2016). A sensitive and specific behavioural screening tool could have been an efficient and cost-effective means of identifying children who may benefit from further assessment (Lange et al., 2017).

There is much debate in the literature regarding the neurobehavioural profile of FASD. It is recognised that there is a considerable challenge associated with identifying a neurodevelopmental profile of FASD. The challenges associated with identifying a neurodevelopmental profile of FASD stems from 1) the range of deficits that individuals with FASD exhibit, and 2) the overlap between FASD and other neurodevelopmental disorders (Lange et al., 2017), and the dynamic nature of cognitive development. Several studies have suggested that while a neurodevelopmental profile can identify children with FASD from typically developing peers (Lange et al., 2019; Mattson et al., 2010; Mattson et al., 2013), the profile is not specific and children with FASD could not be distinguished from other neurodevelopmental conditions such as ADHD and Autism Spectrum Disorders (ASD) on the basis of the shorter test battery (Lange et al., 2019; Mattson et al., 2013). There is considerable overlap in the behavioural presentation of children with FASD and several other neurodevelopmental conditions including ADHD, ASD, learning disability, and conduct disorders (Chasnoff et al., 2015; Weyrauch et al., 2017). Further, given the overlap between experiences of FASD and trauma, children may also present with symptoms related to trauma such as difficulties with behavioural regulation and impaired executive function (Teague, 2013).

While a FASD-specific behavioural phenotype that allows for FASD diagnosis may be unlikely (Lange et al., 2019), identifying the common challenges (i.e., the consistency in presentation) experienced by children with FASD that are relevant to caregivers is

informative for screening. Identifying patterns of challenges, while not specific, could lead families and clinicians to consider the potential for prenatal alcohol exposure and FASD among differential diagnosis. The PFST only includes those domains/behaviours/impairments that were consistently identified in the literature, interviews, and through the expert panel feedback. The domains measured by the items of the PFST include self-regulation, cognitive ability and adaptive behaviour skills. Impairments in these domains - which have been consistently identified as relevant to children with FASD in the chapters of this thesis and in the broader literature (Mattson et al., 2019),- result in behaviours such as difficulties with understanding the consequences of actions, difficulties learning from mistakes, and impulsivity.

Given potential overlaps in presentation with neurodevelopmental difficulties other than FASD (including children who have experienced trauma), it should be noted that the PFST is not designed to conclusively identify FASD. Rather, it should be used to identify children who require further assessment and support. It is recommended that such assessments include assessment of prenatal exposures, as well as exposure to postnatal adversity (such as trauma exposure), particularly if children are in child protection or forensic settings. Examination of the behavioural and cognitive difficulties identified on the PFST may be used to design management and support strategies.

Characteristics of the commonalities of cognitive and behavioural functioning across children with FASD that are relevant to caregivers and clinicians can be informative to screening. While the screening tool may ultimately prove to have a low specificity for FASD when used in population samples due to the overlap in presentation with other neurodevelopmental disorders and among individuals who have experienced trauma, children who need support will be identified and through differential diagnosis assessments will be referred to appropriate treatment and support, including for FASD if PAE is indicated.

Comorbidity of Childhood Trauma and Prenatal Alcohol Exposure

Complex trauma is another exposure that may need to be considered when exploring a differential diagnosis. Children with FASD have been shown to experience high prevalence of abuse and neglect (Kambeitz et al., 2019). Exposure to abuse and neglect can impact children's cognitive and behavioural development in different ways, depending on the type of abuse, timing of abuse and chronicity (Cowell, Cicchetti, Rogosch, & Toth, 2015; Jaffee & Maikovich-Fong, 2011). Trauma is associated with changes to brain structure (Teicher & Samson, 2016) and the symptoms of children who have experienced trauma include emotional and behavioural dysregulation (e.g., lability, anhedonia, explosive or sudden anger), difficulty understanding and expressing emotions, inability to concentrate, disrupted executive functioning, inter-personal difficulties, language impairment, and can include sexualised behaviour and harmful sexualised behaviours (D'Andrea et al., 2012; McLaughlin et al., 2017). There is substantial overlap between some of these symptoms and those symptoms that are displayed by children with FASD. This may be as a result of impairments caused by PAE and child abuse and neglect affecting brain and cognitive development in similar ways. It may also be explained through comorbidity of the two exposures (Kambeitz et al., 2019).

As noted in the literature review, numerous studies comparing cognitive and behavioural outcomes between children with FASD and between groups of heavy PAE have noted high proportions of children in out-of-home care. Among the thirty-one studies of children with FASD, or heavily exposed to alcohol, the proportion of children who were reported to no longer be in the care of their birth parents ranged between 17.5% and 100% of the sample of children with FASD. While several active case ascertainment studies were identified in the review, these studies did not report the proportion of children identified as having FASD who were no longer in the care of their birth parents. Children with FASD have

been reported to experience greater rates of abuse, neglect, and mental health or substance use among family members compared to the general population (Kambeitz et al., 2019; Price, 2019). Previous research has also highlighted the over-representation of children with FASD in child welfare systems (Fuchs et al., 2010; Popova, Lange, Shield, et al., 2019) who could also have experienced abuse and neglect.

The studies conducted in this thesis reflect the over-representation of children who are no longer in the care of their birth parents. These rates of placement into care other than birth parents may indicate that some of the children in these samples had experienced abuse and neglect. Children may have been removed by child protection services at birth due to risk during pregnancy such as heavy PAE, or removed by child protection subsequently for other risks, abuse or neglect. In some cases, extended family may have taken over caring roles for children because of risks to the child. In both the interview study (Chapter 3) and the preliminary reliability and validity study (Chapter 5 and 6), there was a vast over-representation of children who were no longer in the care of their birth parents. In interviews of caregivers with children with FASD (Chapter 2) none of the interview participants were biological carers of children with FASD, and only 7% of the children with FASD in the preliminary reliability and validity studies (Chapter 5 and 6) were in the care of their biological parents. However, as noted above, the overlap between children with FASD and experiences of trauma is not unique to this study.

While questions directly relating to trauma history were not included in the semi-structured interview guides in Chapter 2, or the questionnaire in Chapters 5 and 6, information about experiences of trauma was offered by caregivers unprompted through the course of their participation. Three interview participants (21.4%) spoke to their children as having past experiences of abuse or neglect. Further, one interview participant reported that their child had a comorbid diagnosis of Post-Traumatic Stress Disorder (PTSD). Clinicians

discussed the importance of understanding both PAE and trauma history when conducting diagnostic assessment. Similarly, two (4.8%) caregivers of children with FASD in the preliminary reliability and validity study identified that their children had a comorbid diagnosis of PTSD when they were asked about comorbid health conditions. Further, children with FASD in the preliminary reliability and validity study had high levels of trauma-related symptoms as measured by the BAC-C (FASD = 20.63 (6.94), CON = 4.00 (4.46), $p < .001$) (Tarren-Sweeney, 2013). The scores on the BAC-C among children with FASD were considerably higher than previous studies using the BAC-C, including the BAC-C development sample ($n = 347$, 4 to 11 year old children, Mean BAC-C = 10.0 (8.00)), and a sample of Dutch children in foster care ($n = 118$, 4 to 11 year old children, Mean BAC-C = 12.09 (8.16)). These results suggest a high level of trauma-related behaviours and mental health concerns amongst children with FASD recruited in the preliminary reliability and validity study.

A growing body of research is examining the cognitive and behavioural presentation of children who experience both FASD and trauma. A recent review found exposure to both alcohol prenatally and post-natal abuse and neglect results in poorer cognitive and behavioural outcomes than either exposure alone (Price et al., 2017). Several studies have suggested that exposure to post-natal adverse events increases the risk of poor mental health outcomes among children exposed to alcohol prenatally (Andre, 2019; Ritter, 2019). However, the scarcity of research in this area, and conflicting findings from recent studies highlight the need for further research (Mukherjee et al., 2018; Price, 2019).

It is recommended that FASD screening and assessment efforts are responsive to the high rates of co-occurrence between FASD and trauma and include assessment of pre- and postnatal adversity (including PAE) in developmental history taking. Examination of etiological factors may guide directions on appropriate treatment. This is particularly relevant

for screening among populations considered to be high risk for FASD where there is also high risk for trauma experiences (e.g., children and youth in out of home care, and children in youth justice settings). However, developmental history taking in these populations can be quite difficult as the biological parents may not be involved (Bakhireva et al., 2018). Children and youth in out-of-home care and in youth justice are considered to be at high risk for experiencing FASD (Popova, Lange, Shield, et al., 2019)

Developmental Considerations

The age range of children included in the studies within this thesis (children of preschool and primary school age) represents a period of rapid development. Many of the cognitive and behavioural domains identified across the thesis undergo substantial development through childhood. For example, children learn to regulate their emotions (Morris, Criss, Silk, & Houltberg, 2017) and children's executive functioning undergoes protracted development through childhood (Diamond, 2002). As this age range represents a critical period for development, early identification of children who are experiencing difficulties is a crucial consideration so that early provision of support may prevent or ameliorate long term difficulties (Petrenko et al., 2014). The early identification of children with FASD is, however, difficult. Early identification of some difficulties is compounded by the emerging nature of children's cognitive and behaviour skills. Some skills develop more slowly across childhood (for example, higher order executive functions such as planning) and assessment of young children (i.e., before the age of eight) on these domains is not recommended as these skills are yet to develop or emerge. Further, a large proportion of children show symptoms of hyperactivity and impulsivity as part of their normal development (Campbell et al., 2014)

Across a number of cognitive and behavioural skills domains, differences between children with FASD and those without are likely to become more apparent as children

develop (Tamana et al., 2014; Taylor & Enns, 2017). Related to this, one study reported at age five that only 10% of children with FASD presenting to a diagnostic clinic had significant attention concerns, whereas at age 10 more than 60% had clinically significant attention problems (Astley & Grant, 2012). The preliminary reliability and validity study (Chapter 6) has found that it is possible to distinguish between children who have FASD and those without, however, the accuracy of the PFST in different age groups was unable to be tested due to the sample size. Suggestions for further research in this area are made later in this chapter.

Understanding which behaviours are cause for concern at different ages may enable the creation of tools that are more suited to particular age ranges. As highlighted in the preliminary reliability and validity study, there are some items which may not be applicable to younger children (e.g., “struggles with complex maths problems”). This highlights the need to incorporate response options into the PFST for items which may not be developmentally appropriate for younger children. The literature review found children with FASD experience difficulties in language, academic abilities, some elements of executive functioning, emotional and behavioural regulation, caregiver rated sleep behaviours, and sensory processing at young ages. In the interview, study caregivers were asked about the difficulties that they first noticed in their children. Caregivers identified difficulties with behavioural and emotional regulation as early warning signs.

There are a number of factors which can promote child development as children mature, with parental education levels being strongly associated with children’s behavioural and emotional functioning (Duncan & Magnuson, 2012; Walker et al., 2011). The community sample demonstrated a high level of parental education with almost 90% of the community sample reporting post-graduate education and with such high levels of parental education, the community sample may be performing better than a general population sample. The high

levels of maternal education in the community sample may further exaggerate the differences between the FASD group and the community sample. This difference between groups may also contribute to the high level of sensitivity and specificity found by the tool. Specificity can be falsely raised when the comparison group in diagnostic or screening accuracy studies "is skewed towards greater health or the wellest of the well" (Kohn et al., 2013, p. 1198). Based on the current sample, the specificity and sensitivity reported in this sample may be an over-estimation of the accuracy of the tool. Consequently, further research is necessary on a sample more reflective of the populations where the tool is likely to be most used to determine the validity of the tool. Validation is an on-going process, and although the PFST shows promising preliminary results, future research will be necessary to further examine the validity of the PFST. Future research directions for the PFST are highlighted in greater detail later in this chapter.

Clinical Implications

The development process of the PFST reflects a universal screening process (e.g., the development focused on identifying children who display the cognitive and behavioural characteristics consistent with children who have FASD, from a community sample). With that in mind, the tool should demonstrate a high level of sensitivity to ensure children who display the cognitive and behavioural impairments consistent with FASD are identified and can be referred for follow up assessments. The PFST was developed for universal screening purposes (i.e., the development process has been about identifying children who may have FASD from typically developing peers/community samples), rather than trying to distinguish FASD from other cognitive and behaviour presentations or disorders. It is important that subsequent refinement and development of the PFST are clear on the ways in which the tool is intended to be used.

Universal screening for conditions that are low in prevalence in the general population requires special consideration, given a large number of false positives are likely to be identified. Active case ascertainment studies have estimated the FASD prevalence rate at between 2 and 5% in the general population (Kodituwakku et al., 2006; May et al., 2018; Popova, Lange, Poznyak, et al., 2019), and while higher estimates have been reported for Australian subpopulation samples, these are not likely to be representative of FASD Prevalence rates in the general Australian population. In low prevalence situations, identifying individuals who may have the condition from those who do not have the condition is a key consideration (Streiner, 2003). To this end, tools that displaying a high sensitivity and high negative predictive value are ideal properties of a "rule-out" test (Florkowski, 2008). As illustrated by the results in chapter 6, assuming a populations prevalence of 2%, the PPP of the PFST was 0.214, indicating that 78.6% of positive tests would be false positives.

In many situations where screening is conducted for low prevalence disorders, an initial test with high sensitivity is followed up with a second test with higher specificity before conducting any expensive or potentially invasive diagnostic test (Evans et al., 2005; Streiner, 2003). Caregivers have previously described proxy-report measures and screening tools as an opportunity to "flag up" behaviour concerns, and as helping clinicians to think in a "sharper, more focused way" (Frogley et al., 2019). The outcome of a positive screen on the PFST should prompt referrals for further assessment and ensure questions around alcohol use in pregnancy are asked alongside other prenatal and postnatal exposures. Further, given the increased likelihood that children with FASD may be over-represented in samples of children who have experienced abuse and neglect (and potential removal from birth parents) (Kambeitz et al., 2019), it is recommended that a complete developmental history include PAE and abuse and trauma exposure accompany assessment. Tools focused on identifying trauma-related mental health difficulties such as the Assessment Checklist for Children, or

the Brief Assessment Checklist for Children (Tarren-Sweeney, 2007, 2013) could potentially assess for comorbidity given it includes abuse-specific items (e.g. sexualised behaviour, other examples). An approach of screening and follow up assessments seeking information about PAE has been suggested (Weyrauch et al., 2017). Subsequent assessments should be geared towards informing intervention practices and ensuring that the interventions proposed are appropriate for meeting the child's needs (Pei et al., 2013). Further assessment can prove valuable without a FASD diagnosis, as it may still provide information regarding the nature of children's difficulties and can inform further diagnosis or interventions (Pei et al., 2017).

A salient challenge to further testing of the PFST is that all individuals participating should receive follow up assessment as part of the study to determine whether participants have FASD. As mentioned in the introduction, the cost of diagnostic assessments for FASD are prohibitively high if done at scale (Mukherjee et al., 2018; Popova et al., 2015). The screening tool may also be used in a tiered or simultaneous process wherein children who screen positive on the screening tool are referred for a second, more specific test. Tests that are more difficult to adopt at scale may be considered for the second tier including developmental history taking to guide next steps (e.g., exposure to alcohol consumption and/or exposure to trauma), dysmorphological assessments (Astley et al., 2002; Suttie et al., 2017), neurocognitive testing (Lange et al., 2019; Mattson et al., 2013), or eye-tracking (Zhang et al., 2019).

Physical features were not considered in the development of this screening tool. Parent/caregiver report of behaviour was selected as the target for this screening tool as the cognitive and behavioural difficulties of children are likely to be the cause of concern for parents and caregivers. Several effective methods for physical feature screening already exist including the use of digital photography (Astley et al., 2002) and 3-D facial imaging Suttie et al., 2017). These methods may be employed alongside a tool such as this in a simultaneous

screening approach. For example, future research may include the application of physical features screening and the PFST to determine if there is an increase in accurately identifying children with PAE. Composite screening methods have been explored in previous students such as Zhang et al. (2019) who investigated a composite screening procedure involving eye tracking, neuropsychological testing and brain scans.

Study Limitations

Several study limitations have previously been highlighted and discussed throughout the thesis. In this section, three overarching limitations that should be addressed in future research examining the PFST are discussed. These include recruitment methods for the FASD and community samples, the gold standard against which the tool was judged, and the small number of clinicians who participated in the study.

Recruitment of carers of children with FASD occurred primarily through social media and mailing lists of organisations established to support children and families living with FASD. While demographic characteristics of the membership of these groups was unavailable, the accounts are likely to have two main audiences: 1) those raising a child with FASD who are actively seeking information and support, and 2) those who have a professional interest in FASD (i.e., health, allied health, social work, research). In addition to the organisations through which participants were recruited, the recruitment made no attempts to disguise the purpose of the research, and the studies clearly identified the objective of the research to develop a screening tool to identify children who may have FASD. Both the sampling methods and the advertisement of the studies may have introduced potential bias into the sampling in both the FASD and community samples. Four potential sources of bias for both participation in support groups and participation in the study are discussed below: (1) lack of awareness of FASD, (2) lack of relevance, (3) stigma and guilt, and (4) difficulties in participation in the research among birth families. It is important to note

that these sources of bias are not unique to this study. Firstly, families who do not know that their child has FASD or clinicians who are unaware that their clients may have FASD would have been excluded from the research on the basis of the recruitment strategies. Secondly, a number of participants in the community sample may have thought that participation was not relevant to them, given the explicit focus on FASD. The final two points relate primarily to biological parents of children with FASD. Difficulties in recruiting children who live with one or both of their biological parents may be related to stigma associated with FASD (Choate & Badry, 2019; Corrigan et al., 2017), the parents' feelings of guilt and grief regarding having consumed alcohol during pregnancy (Domeij et al., 2018). Further, among children placed into out of home care, kinship carers may be more reluctant to seek support compared to non-biological foster parents (Coleman & Wu, 2016). Lastly, biological families living with FASD may experience a number of barriers to participation in support groups and research due to social and economic disadvantage. A review of the characteristics of biological families of children with FASD highlighted a number of risk factors, and high levels of disadvantage (Esper & Furtado, 2014). Further, for participants who are in active addiction, participants may be especially difficult (Thomson, Morley, Teesson, Sannibale, & Haber, 2008). Biological parents of children with FASD may be considered a hard to reach population in research owing to the social disadvantage identified, and if there is potential involvement in a child protection case (Mirick, 2016). Overcoming these barriers may require highly targeted recruitment strategies, such as through drug and alcohol treatment clinics, hospitals, paediatricians and family support services (Bonevski et al., 2014). Building screening tools into practice, for example including the PFST as a cognitive and behavioural screening tool as a part of standardised assessment process, in hospital, school or treatment settings may reduce the stigma and potential bias in responses (Eapen & Jairam, 2009).

As noted, sampling bias in the community sample may have contributed to the results of the preliminary reliability and validity study. The community sample included a high prevalence of individuals with postgraduate education with approximately 90% of the community sample reporting post-graduate education. The high level of maternal education present in the community sample limits the conclusions that can be drawn about the screening tool and highlights the need for further studies in a broader sample, more representative of the populations that may seek assessment for behavioural concerns. The recruitment strategy may have contributed to this bias. Convenience samples have been shown to recruit individuals who feel strongly about the issue in question, and the community sample may have been drawn from professionals who subscribe to FASD support groups.

A further limitation in this PhD study was the "gold standard" against which the screening tool was judged. In the current studies, the FASD diagnostic group was comprised of caregivers who reported their children either had a FASD diagnosis or were suspected of having FASD. Current best practice for the diagnosis of FASD is for a multidisciplinary team assessment to be conducted, incorporating assessment of all the neurodevelopment domains (Bower & Elliott, 2016). Caregivers in the qualitative study (Chapter 3) and the preliminary reliability and validity study (Chapters 5 and 6) reported their FASD diagnosis was obtained using a range of approaches including multidisciplinary assessment, to diagnoses formulated by a single clinician. In addition to the variability in who conferred a diagnosis, there may be variability in the criteria against which children were judged. Previous research has shown variability in outcomes if different diagnostic criteria as applied to the same cases (Coles et al., 2016). This is a particular concern for children diagnosed prior to 2016 before the introduction of the Australian Diagnostic guidelines (Bower & Elliott, 2016). However, the diagnosis of FASD in Australia is typically undertaken by a small number of expert

clinician/clinics across the country (Zimmet & APSU FASD Investigators, 2019), which may balance against this variability.

A recurring limitation through the earlier stages of the development of the PFST was the limited participation by clinicians. Despite numerous attempts to recruit clinicians into the interview studies as highlighted in Chapter 3B, only a small number of clinicians participated in the interviews. Further, while all clinicians consented to take part in the expert panel study, responses were only received by two clinicians, despite follow up. There may a range of factors which contributed to this including limitations of the recruitment strategy and time commitments associated with participation in the research including a small pool of expert clinicians who are currently involved in the diagnosis of FASD (Zimmet & APSU FASD Investigators, 2019). While there are, however, extensive efforts underway to train a wide range of clinicians about FASD (Reid, White, et al., 2019), the valuable contribution of the small number of clinicians who were able to participate, as evidenced in Chapters 3 and 4, highlight a need for further work to validate the PFST among clinicians. Several avenues for enhancing clinician participation and further refining the PFST could be explored. The feasibility of the tool could be examined in settings such as a hospital or school. Further, clinicians could be recruited through clinical networks to further refine the tool.

Future Research Directions

The suggestions for future research detailed below are in addition to those mentioned in the preceding Chapter discussions which have been highlighted throughout the thesis. Further testing of the PFST in a representative sample, for example through preschools and schools in conjunction with an ACA process, is required to address the biases noted in the present study. Recruitment processes could be modified in several ways to ensure greater participation and reduce sampling bias. Modifications may include not identifying the tool as relating to FASD to reduce any potential bias relating to stigma associated with alcohol

consumption during pregnancy (Choate & Badry, 2019; Corrigan et al., 2017) or the perceived relevance of the tool, or the PFST could be embedded into standardised assessment. These modifications, combined with the more targeted approaches as outlined above, may be considered to encourage biological parents to participate.

Future studies should be conducted to refine the number of items retained in the tool. The item refinement conducted to date was based on a relatively small pilot study, with some bias in the samples as discussed. Future research with a larger, more diverse sample may reduce the item list further, however, accuracy should be prioritised over brevity. Factor analysis was not undertaken in this study due to the small sample size (Reio & Shuck, 2015) and future studies with larger, more representative samples, may examine the factor structure of the tool to provide further evidence of the psychometric properties of the PFST. For future research, a three factor structure examining the over-arching domains of the tool and consistent with the Neurodevelopment Disability associated with PAE criteria (American Psychological Association, 2013) could be investigated. While research across countries would enable access to a large pool of participants to enable factor analytical studies, research among samples outside of Australia may also require cross-cultural validation studies (Peña, 2007).

Further examination of whether the tool performs equally across age ranges is required. The transition to school has previously been identified as a time when behavioural and cognitive concerns become more apparent. While no relationship between the PFST and age was noted in this study, future research could examine the screening performance of the tool across age groups. The predictive accuracy of a tool such as the PFST at successive age bands could be explored to examine the earliest ages at which the PFST may have accuracy as a universal screening tool. Examination of the accuracy across age bands may have implications for age-related standard scores. Methods of analysis based on Item Response

Theory such as Rasch analysis may offer further insights into the application of the PFST across age ranges. Lastly, as noted above, some of the items may not be applicable to the youngest age group who are not yet attending school (e.g., struggling with complex and simple mathematics). A “Not applicable”, or other more developmentally appropriate response option should be added for all questions.

There needs to be consideration as to whether the screening tool should include prenatal alcohol exposure questions. Embedding a brief alcohol exposure questionnaire into the screening tool may help in distinguishing FASD from other conditions that are not a result of PAE. However, the sensitive nature of a questionnaire relating to alcohol use in pregnancy may discourage some participants. Future research could explore the feasibility of embedding these questions into the PFST, considering how best to ask these questions (Muggli et al., 2015).

Conclusion

This thesis sought to develop a screening tool to identify children who display the cognitive and behavioural difficulties consistent with FASD. Through a series of sequential stages, the existing literature and caregiver and clinician perspectives were included in the development of a screening tool to identify children who display cognitive and behavioural impairments consistent with FASD. A broad range of cognitive and behaviour domains were identified through the scoping review and interviews which were important for any tool intended to capture the behavioural and cognitive profile of FASD to measure. The 57-item PFST demonstrated a high level of accuracy in identifying children who are at-risk of having FASD from a community comparison. There were, however, several sources of bias identified in the pilot study which future research will need to address. This thesis presents the initial steps in the development of a FASD screening tool, and there are additional steps to be taken before it can be determined whether the PFST could be applied at a whole of population level

or within targeted samples. Despite the limitations of the study, this research offers a promising FASD screening tool that with further refinement may enable children who display cognitive and behavioural impairments consistent with FASD to be identified early, to receive referrals to diagnostic and assessment services, and to receive tailored interventions that best support children with FASD and their families.

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Appendices

Appendix A: Additional Caregivers Interview Extracts

Subtheme	Exemplar quotes
Major theme: Regulation (Behavioural	
Hyperactivity	The teacher would have to let him walk up and down the balcony, or run up and down the balcony through the day just to release energy. He couldn't sit still. [P1] Yeah, look, I suppose with our youngest he is very fidgety because he has ADHD. [P2] And honestly, he was always just off the show. He really was. And at the time, my three girls were at school and gender differences as well- I hadn't had a boy so I could never quite tell- "is this boy behaviour? Or is he odd?" you know. But he was always just so wired and hard to control. [P3] Always on the go. Always. She can't sit still. Actually, that's the real trouble they have had with her at school- getting her to stay in one spot. [P4] Well, when I first got the oldest one- I had no idea fetal alcohol was a thing, I had no idea. He was very, very, very, very, hyperactive [P6] One of the other things I just thought of that is a problem day to day for him is hyperactivity for him as well. [P7] From the time he gets up until he has his medication and calms down, yeah- it's like a tornado hitting the house. He just races around everywhere and turns everything upside down [P9] And he's becoming a lot more hyperactive, makes constant noises, just constantly, he gets into trouble all the time at school because he makes just constant noises. He's disrupting the class, and he doesn't even know- he has no idea he's doing it. [P10] We can't get her to sit still long enough to actually listen to a story. [P11] and they're very active and… what's the word for it? I can't think of the word… I don't know, they are just too full on…[P12] No ability to sit still, or to stop talking. There is white noise coming out of her the whole day long. From the moment she wakes up to the moment she goes to sleep. Just white noise all the time. So, whether it's (makes humming noise) or just continually, continually noise. And she cannot sit still. [P13] But a high level of ADHD. So we don't think that he can turn off very well. And when he is awake, he is just straight back into that. He's either on or off. And when he is on- he's everywhere man [P14]
Impulsivity	One day he had this big plastic tube and he was actually sitting next to me, and he was on the lounge chair eating… well he was supposed to be eating, well I was watching him, and the next minute he starts coughing and I said "what are you doing?" and he's got off the chair and he… and I looked over and he was really coughing and what he had done, he had got this plastic tube- I don't even know where he got it- and there was all this glitter and he breathed up the glitter and I said "why, why would you do that?" And he goes "oh I don't know, I just wanted to suck it up". [P1]

Subtheme	Exemplar quotes
	Because he (8 year old) is so impulsive socially, he's probably going to be getting himself into strife. His mates know that if they ask him to do something that he will do it [P2]
Impulsivity	She's very impulsive. [P4] I don't know whether this is a behavioural thing related to the FASD but she has a tendency to take things that don't belong to her. [P5] Like if there is a scratch on the wall, well next minute there is a hole in wall because he scratched it to see what's behind the scratch. [P6] my son actually has a high IQ… which means that he has got really good ideas but he's got no buffer about them being safe or whether they can or can't happen. [P7] I think like an impulse thing. He has no idea like that doesn't belong to me, I shouldn't touch it, or I've been told not to touch it. [P8] Oh yeah, he doesn't stop and think about anything. Yeah, he's… he's full on. If he sees something and wants to do it, he'll give it a go, mate. He doesn't think about what the consequences are going to be. [P9] I don't know how to explain it… he can just, he's quite impulsive [P10] Even when we are out going shopping, as she will just wander off, climb over shelving- anything- and carparks are particular bad. She still is not getting that we have to wait and look before we cross roads. [P11] You ask them to clean their room, you are going to end up with odd shoes, clothes sets that are missing bits, their toys… everything will get thrown out. They just don't care. [P12] you know, they might be playing hide and seek in the house and she'll just "Okay, the shower bay is a great place to hide" but she just wouldn't think, and she'd just turn the shower on [laughing]. She's full clothed right. And we are playing… Okay. Or she'll go visiting at friend's house- "Oh that morning fresh looks nice in that container, I'll just tip that out into the sink". The full 750ml… [P13] I've described it as the monkey driving the bus… and I've also given the analogy to people that his thinking processes are slow like Winnie the Pooh in a Tigger body. [P14]
Risk taking behaviours	But he takes a lot of dangerous risks, he doesn't think about the danger of what he is doing. [P1] We had a case where we were at the snow, and he asked what this great big red sign was and, you know, it was to not sit somewhere because the snow could tumble off the roof of this building. And he could see that it was something like that, he could see it was some kind of warning sign. So I explained it to him, and that was fine. And when I went to pay for our lunch, we went and had lunch and I paid for lunch, he just bolted, he just took off while my credit card was going through the machine so I couldn't just grab him and it was sheer panic. There was people everywhere. And had no idea where he had gone, and then it just clicked, he would be sitting under that sign. And that's exactly where he was. So, he would want… yeah he would want to see what happens. So, if there is any… anything like that that would look dangerous, he would want to go and find out what would happen if he did that. Absolutely, without a second thought. That's just one example that's stuck in my head where it was worked to my advantage that I knew where he would be. You know where was the most dangerous place and that's where he will be. [P2] She's got no sense of danger really. I've known her to throw things out windows and jump out things, or off things. [P4]

Subtheme	Exemplar quotes
	He would be awake then until 3 or 4 in the morning, into things, like destroying the kitchen. He burnt the kitchen down when he was about four. We call him the stealthy chef, because he'd be so quiet we wouldn't hear him wake up and he'd be in the kitchen destroying it. [P6]
	And a lot of the things like he will do on the playground that are more adventurous would be because he is more risk-taking. [P7] Oh, with the push-bike trying to jump… like he set up this great, big jump trying to jump things and climbing on top of the shed and thinks he's going to jump off and yeah. Because he sees things on TV or hears someone talk about doing these things and yeah, he'll want to try and do it himself. [P9] I've had them up on the roof. We live in a [homestead?] house, and we've found them up on the roof. They climb play equipment at the park or at school- they don't use them how it's intended. They climb where they are not supposed to be and play there. They climb fences in dangerous places and run out on roads. [P12] He can bound across the road before he realises that there is a car coming, or that it's not a great idea. [P14]
Taking things which don't belong to them	The younger one, he steals all of the time. He shoplifts all of the time. And even, when we will say…. Because he loves Bunnings, and it'll be things like washers, and screws, and padlocks. Anything metal, chains, bits of chain. Like we'll have a talk before we go you know, "You can't take anything with you. Do you know what could happen if you got caught for shop lifting?" and all this. And we pat him down every time and there will always be something. [P2] He has particularly light fingers; in the sense that he has this… he's a great little thief. I think like an impulse thing. He has no idea like that does not belong to me, I should not touch it, or I've been told not to touch it. [P3] Actually, that's the other thing with the impulsivity and stuff. We have trouble with shoplifting and lying. [P4] I don't know whether this is a behavioural thing related to the FASD, but she has a tendency to take things that don't belong to her. [P5] The oldest one- we wouldn't call it stealing because he doesn't have intent to steal it. He just takes things. He doesn't understand what's mine is mine and not his. [P6] We are very careful if we take him into shops. There has… we have to check his pockets actually before we go anywhere or leave anywhere. There have been some things that have appeared at home that I can't recall having bought them for him. [P7] [In response to interviewer prompt about taking things that don't belong to them] – [laughter] Yes, been there, done that one. [P9] No, I've never had that, nup none of that. [P10] She will, however, fill my trolley but no, no stealing as yet. [P11]
//	We've had food stealing as well- from other children at school. Despite having similar things in our own lunch boxes, it just looks more appealing. And it is spur of the moment- I'll have that, I want that". They don't realise what that means for the other child… they have no concept. [P12] Yes, yeah absolutely. Yeah, that's just normal. It's just a part of our lives. I do check her pockets and stuff afterwards. And the amount of times that she's had to go back and pay for stuff- and I make her do that. There's no point doing it the next day, there's no point doing it a week later… [P13]
Major theme: Regulation (Emotional)	

Subtheme	Exemplar quotes
Tantrums or meltdowns	A lot of breakdowns and meltdowns. [P1] Even at this age of 10, he's still very much a tantrum-based child, like I can't have that so he has a hissy fit, a very infantile response like a (crying sounds) like a, you know just really inappropriate for that age. [P3] He has a lot of anxiety, so he is really attached to his teacher and if his teacher has to go to the toilet or go to lunch, he can't handle that he breaks down and has a meltdown. [P6] It's like he's on a high all day. He's very explosive- the tiniest little thing will set him off. [P7] We go into the massive meltdown because she is not getting her way. [P8] It could be anything. He could be sitting there, playing around, talking not a problem, and away he goes. It's like living with a stick of dynamite mate; you never know when it's going to go off. [P9] Yeah, yeah, he does. [Have tantrums often] He'll scream and… [P10] I can remember one morning I said to one of the them, can you put on this pair of shoes please, and she has gone "Oh I want those shoes' and I said "Well they are not appropriate for today and you need to wear these ones", or something like this, and it resulted in a twelve-hour meltdown. [P12] Or if she didn't get what she wanted, she did over the top meltdowns. [P13] Yeah, he's very reactive- the tantruming, reactive, the crying, he's so worked up you can't go to sleep. [P14]
Managing transitions	Yeah, it is not there at all. He, umm, and I think like a lot of kids he loves the computer, he loves playing with, he loves playing Minecraft and once again like a normal child, it's very hard to get him off. We've tried all of the strategies of you know, here's a timer, you know, look at the clock, and when the big hand is on this time you are coming off and, we are not going to have a meltdown because you know you are coming off but it always happens. [P3] And when those sorts of things happen, it's not just a spur of the moment thing that I say "ok we are going inside" now. There would have been warnings, there would have been an explanation before we go outside for how long we are going to be out there for, what we are going to be doing, and that when I'm finished doing what I'm doing, then we will be coming inside. And what he will do is he will say yes I understand, yes I know. Yet… although he is saying the words… he's not and yet if I ask him to repeat that back to me, he will become very angry. [P7] A couple of times when I've gone to pick her up, she's had meltdowns because I don't know, she hates the transition period. And I have noticed the girls are completely confused by what she is doing, and have no idea what, why, or anything, but she gets quite angry. [P11] I think she had a lot of trouble transition from one thing to another. Like huge, I'm talking… moving from playdough to paint, she would just throw herself down, or have a great big tantrum… [P13] You need a lot of time if you need to change something. There has to be a lot of talk ahead of time- so for example if we have to get up early to go and do something else first before you do the kindergarten drop off, you have to tell him in advance who is being dropped off first. And often

Subtheme	Exemplar quotes
	he screams the whole way, he wants to be dropped off first- it does not fit with the program from the day. [P14]
Major theme: Regulation (Attention)	
Easily distracted	Well now that he's on Ritalin, it's a lot better cause he has that in the morning. It actually makes him focus and concentrate and actually achieve a task even though he's not doing the same level as all the other kids. He actually sits there and completes something even if he's a lot slower at it. [P1]
	Focusing is difficult for him [P3] She has trouble with a lot of things at school. And she can't sit with the other children otherwise her concentrate level just goes. She even told the psychologist that, so that was interesting. She came out with that one herself, and we just sort of went "Ohhh, okay". [P4] And she could come out a couple of times afterwards, you know what I mean, so she's easily distracted. So that's one thing as well. I think that plays on the short-term memory thing for her. [P5] And it is a matter of "Put your shirt on. Put your pants on". You know, like you said, one thing at a time. [P6] Oh absolutely, I mean, look, not even half through the exercise, it could be almost instantaneous. So, she's very easily distracted and I think that's what the issue is. [P8] She'll quite happily draw a person, but then that's it- "Oh sorry, something's distracted me" and off she goes. [P11] Inattentiveness [P12] She gets side-tracked very easily…[P13] Easily distracted. [P14]
Needing external support	Because she can't focus, and she knows she can get away with just drifting off because somebody is not watching her all of the time. She just gets lost in her little world, and the teacher will bring her back to reality. [P5] Something you've got to just keep reminding her because there is that short term memory. Like she'll be standing there, and I'll go what are you doing- ohhhh, ummm- "weren't you doing this?", "Oh yeah, that's right mum"- off you go, go and get it done. [P8] Have to watch her constantly. like a toddler. Because if you leave for a few minutes, she just disappears or is into something or doing something that she shouldn't be doing. That's the same anywhere, if I go to any sporting… go out anywhere, he's always gotta with you because if he gets out by himself, well, he'll just take off and go off and yeah. So it's full time, twenty four-seven keeping an eye on him [P9] They have trouble being focused. You've got to prompt them and remind them. I don't know… just constantly. But they just side-track so easily, and just start doing something else. You can't keep them where they are meant to be. It's a constant struggle. [P12]
Major theme: Cognitive Ability (Memory)	
Difficulty understanding consequences	Not understanding consequences. [P1] Yep, he just doesn't get it at all that there is consequences. [P2] Yeah, yeah… he's very trying. SO even when you do something fun, or you know you go out for the day or do something, he still can't

Subtheme	Exemplar quotes
	connect- if I'm polite, if I'm quiet, if I do the right thing, like if we are going on a long drive, he doesn't correspond that I do all these good things, dad will probably give me his phone and let me play on the phone for half an hour. He can't think "good behaviour gets good rewards". So, he can actually… he spoils a lot of events for himself because he doesn't click that… I'm actually making this difficult for myself. [P3]
	She doesn't understand consequences for actions very well. So yeah, and if you put her in time out for something, it can't be more than five minutes otherwise she forgets why she is there. [P4] But she'll lie about almost anything but she doesn't understand consequence- you know? [P5] The word consequences is a really, it's a really bad word for him. I use good effects and bad effects to try to sort of bring it all in that way but even with that, umm, knowing that there is bad effects that will happen because of things- and I will be warning him, I will say "If you continue this, there will be a bad effect for it" and that, that does nothing for him. [P7] yes, she understands. She understands. I said she'll just scream and carry on and no whatever, but once she calms down then we will sit and talk, and I go "Do you understand why I did that?", "Yes mum, because I was being naughty", and I said "when you are naughty what happens?", "I don't get to do nice things" and I said "Correct". [P8] Nah, no not really. He can do the same thing, you know, twenty times a day and he still doesn't realise what he is doing wrong. He always will tend to blame whoever is around- he'll blame them for him behaviour like that. But yeah, time out and all those sorts of things don't work. We still sort of try it, none of it works, none of those sorts of things work. Any sort of- taking things off him for his behaviours- none of that works either. You could take everything off him, it doesn't… it wouldn't matter. [P10] She will not understand the consequence - if you throw something, it will be taken away, if you hit somebody you will be removed- stuff like that she still will not get it. We try to restrict anything that excludes her because she cannot understand if she is being excluded for any reason… but for the most part, it's generally removal of an item. But if the consequence is too long, and not immediate, we can't. She won't get it. So, it has to be an immediate consequence. [P11] P: Don't even bother! [Disciplining] Because it has no impact- like "yeah whatever, ok whatever". [P12] P2. they can't put them together… if you have to discipline them in anyway, they don't understand… they haven't got the connection… P1: …with their action… P2: between what they have done to what the consequence could be. P1: Yeah, yeah, that's right. P2: So it makes it very difficult to be able to discipline them in that sense. [P13] He can't be corrected, you can't ask for good behaviour, he just can't connect what he's doing with reward, he doesn't connect what he is doing with consequences or outcomes. [P14]
Working or short-term memory	But then he'll forgot five minutes later and he'll turn around and repeat exactly the same thing. [P1]

Subtheme	Exemplar quotes
	He's got a poor working memory, so do you genuinely not remember what I told you, or is it really a blank? [P3] You can't give her more than one instruction at once. You can't say "brush your teeth, put your pajamas on, go to the toilet", or whatever order you give it in. You have to give her one "brush your teeth" and then you have to go and make sure she's actually doing it, because if she goes out into that hallway and sees something else, or suddenly remembers something else she wants to do, she's gone and you can't… you have to chase her and say "Right, get back here, come back and do this, what were you just told to do?" [P4]
	Look, its… its… you know… she struggles with of course, memory, you know. Short term memory more so. [P5] Her short-term memory is very very poor, very poor. [P6] So, it's sort of a responsive on very clear directions so no more… she's up to probably about three responses that she can deal with in instructions. [P8] And she can, you know, if you ask her "can you go and put your bag away?" and she gets halfway down the hallway and she doesn't know- what am I meant to do again. Sort of more memory type things, she doesn't remember a lot of things. [P10] Definitely, definitely. You can't say three things at once because they'd never get any of that. You've got to go "Go and brush your teeth, go brush your teeth, have you brushed your teeth? Why aren't your teeth brushed?" you can't go "get ready for school"- that's just too complex. You have to go "where is your shirt? why don't you have your shirt on? Can you please put your shirt on? It's mind-boggling, it's mind numbing. [P12] I think sort term memory is a really huge one. So, ability to remember a verse that our six year old read on the previous page, it's just not there- that ability is just not there. Or the ability to remember what is happening in play right now socially. Or the ability to remember multiple instructions. That ability is just not there. [P13] So swimming is taking a bit longer… then we would have liked, but he is on target for a child that struggles to remember the instructions. [P14]
Forgetting previously learnt information	Or being told something and then just staring at you like they have never heard that before "you've never said that before" and it could be yeah something pretty major. [P2] He's (6-year-old boy) expected to learn his sight words, we've been on the same twelve sight words since the beginning of last year. And every night we can read them and by the fifth time we've read them he can tell them to me but by the next day he has no idea. Whereas other children are on number 200, we are still on the first twelve" [P6] And today they might know something like you might run through the spelling words with them from school, and they know them and then five minutes later when you try to go through them again, they've got no idea. No idea in the word, because it's just gone. [P12] Even though you think that he should know, you just can't ever… and then he gets really angry if you, like, try to hold his hand when he knows that he shouldn't, you know… it's almost like a can't really win situation sometimes. If you are silly enough to think that he has recalled it this time… there is a good chance that he hasn't. [P14]

Subtheme	Exemplar quotes
Need for repeated exposure	It's the continuous repeating of every single thing. [P1]
	Just not learning at the right pace yeah. And going over and over and over things and just getting nowhere. [P2]
	There is a lot of stuff that they have to do repetitively with her. [P4]
	It is sort of, I think, because of her short term memory loss, it seems you have just got to keep implementing. [P8]
	It's just routine and repetitive- every day is a new day. You've got to go through the routine, the same thing every day. [P9]
	Yeah, it's just… I suppose saying the same things over and over to him, about his behaviour. You need to… yeah… just that repetitious sort of stuff, you've got to keep saying the same thing over and over again, and hopefully… and at some stage, one day they might click. [P10]
	My niece does always push the limits, like she says "I wanna be dropped off first" and she knows she's always dropped off second. And every single morning it will be the same thing, just like every single morning she'll ask who is going where, even though it doesn't change really. [P11]
	P2: I think it's part of the condition, she doesn't sort of …. Toys, maybe a toy in the cupboard. If it's put back in there.
	P1: If it's put back in the same place a thousand times or more, she might be able to remember where it is. [P13]
Major theme: Cognitive Ability (Academic)	
Academic achievement generally	He's in year five this year and is only doing year two work [P1]
	[Struggles with] Academics. [P2]
	And he likes experimenting, and he likes the science-y sort of factual things. I mean, he's certainly not going to be an academic student, because I don't think he's got the concentration levels to be able to do it. [P3]
	And my partner who is a teacher, like I said, had actually said to me way before that "(Interviewee), I don't think… she's not operating at the level that she should be" and I said "Oh nah, I think she is fine, I don't know the school hasn't really mentioned anything" and that not long after that they started to tell me. [P4]
	She's (11-year old) probably anywhere between year one and three, so they are giving her work content between one and three. Very little year three stuff. [P5]
	And the youngest one is in grade one but he is still doing prep curriculum as well- he's good at religion. [P6]
	And you mention before that he struggles quite a bit at school, does he struggle with most of his subjects?
	P: Yes, he does, yeah. Like, if it's something that he's interested in then he will comply a bit better but if it's something that he thinks he's not going to be able to do he'll either scribble all over the page, he'll just… he'll just tend to do things that will distract the teacher from getting him to do his work because he knows he can't do it. [P10]
	None. We are getting some writing in the form of tiny little lines, I've noticed she's trying to write the letter H which I've noticed because my son's name is (redacted) so she's copying his writing. [P11]
	I: Does she struggle academically?
	P: Very much so. Very much so. [P12]

Subtheme	Exemplar quotes
	I: Yeah, yeah. I was just about to ask how's she going in school. P1: yeah, she's not going so well. She's making progress but not fantastically. [P13]
Maths difficulties	Because he is blooming in maths at the year 2 level, now having a calculator he absolutely loves it. Where if he was to do the grade 5 level, there's no way in hell he would be able to do that. He would hate the subject, he would hate any of school I think. [P1] Maths is a real struggle, all mathematical concepts. They are not too bad when they cook with him but again you've got visual cues and you are discussing with them you know two-thirds what it's going to look like if we cut this cut into three pieces and where we went to fill up to but if that was a sum on a piece of paper, they wouldn't be able to picture that at all. [P2] I think his maths is probably grade 4 or grade 3 [P3]
	she's not very good at maths. She is having trouble with groups of ten. Well, she was of late last year, and I assume she is not much different now. They were just starting to get her into that. She get's very confused with numbers. I can't remember what else they have said, but she really struggles with maths in general. [P4] [In response to question about challenges] Education would be, most particularly Maths. [P7] how is she going to school? Does she, how does she-? P: Fine, I find, I think she's very astute with mathematics. [P8] (male child) has big trouble with the maths. [P9] Struggles at school with maths, writing. [P10] Numbers are much the same as well- she can count, we are getting up to almost fifteen but she cannot recognise numbers and won't attempt to write them. [P11]
Literacy	So, he has a lot of problems…. His writing looks like a prep writing still and he's in year five. He's actually doing year two work. So, his writing is still like a prep grade one-r. [P1] And say with that in particular, if he can copy off the board it's not too bad. But if he has to think what he is going to write, and not be able to copy it, and have to put it on paper then there is huge difference. Like he will forget capital letters, punctuation, the size of the letters, to stay on the line. He needs a lot of cues where it's hard to process several things at once. [P2] He's a very good reader, he's very articulate, so his reading level…English is definitely his strength [P3] So she's… quite a few levels behind in her reading. She is probably… now I'm not sure for this year because they haven't spoken to them yet, because they have changed the way they do some of the parent teacher things with kids who are on the independent learning program. But last year she was in grade two, and she was only at a prep level reading. [P4] The ten-year-old is really doing prep which is five or six-year-old language and literacy level. [P6] Sometimes, it's getting… the frustration comes from getting the word from his head on to the paper and things, he just can't get the letters

Subtheme	Exemplar quotes
	out from his head of what it should be on to the paper. It can't come out the same. So that's really frustrating for him. [P7] Struggles at school with maths, writing. [P10] We are getting some writing in the form of tiny little lines, I've noticed she's trying to write the letter H which I've noticed because my son's name is (redacted) so she's copying his writing. [P11] She was obsessed with books, but she's not taking on board the concepts. [P12]
Major theme: Adaptive Functioning (Communication and Language)	
Receptive language difficulties	He doesn't have a sense of humour, unless it's funny to him, if we say something that's joking, he can't see the fun in it and he takes things as a personal attack, even if you are not particularly talking about him or to him. He just doesn't understand that sense of humour, or the funny side in things like that. [P1] He was having to write something out of a newspaper and you know it's like, "who the article was about, what happened, where it was". It was only two lines, and he said "No, it's not in there, it's not in there" and all the information was in that two lines. So, I re-read those two lines out of the newspaper and he went "Ohh"- but he couldn't read that and hold that information but when I read it to him, he could hold that information for long enough to write it down. [P2] We keep it really simple. And it is a matter of "Put your shirt on. Put your pants on". You know, like you said, one thing at a time.[P6] He can repeat back to me exactly what I've said. But to actually have comprehended that that is what's to happen, that doesn't happen frequently. [P7] He's becoming harder to reason with and just doesn't really understand a lot of things that you are talking about [P10] So we have to really simplify sentences. She can pass every language assessment in the world but it's not going to change it- she can't understand what we say. Then… yeah (laughter). There is a very big lack of communication. [P11] She will copy down books, give her an iPad and she'll start copying a book into her notes, but you ask her what the book is about, and she's got really no clue. She can recall like the names of the characters, but that child had a balloon and the balloon flew away- what colour was the balloon? Oh, I dunno. You've just written yellow balloon four times; how do you not know? [P12] You know, behaviour wise, like he… umm… it was almost like he didn't understand us when we spoke to him, so his receptive language was almost absent. [P14]
Willfully defiant or not understanding?	One thing I know with him too is he comes and he will tell me what he has done because he doesn't think of the consequences of what… like for example. He not long ago smashed a window. He was fighting with my daughter and he just was outside, she annoyed him so much, he just picked up this rock and he just smashed her window and he ran to me and he goes " Mum, I smashed (daughter) window" and I said what, why?, and he said "well she made me mad, I just smashed the window" and, so he comes and he tells you exactly what he has done wrong and that's one

Subtheme	Exemplar quotes
	thing that I've found with him but I've heard some parents say that their kids lies, but I do know that he doesn't lie.[P1] It's hard for us to determine in a situation "are you being defiant, or deliberately… or you can't understand what I'm saying", or do you… he's got a poor working memory, so do you genuinely not remember what I told you, or is it really a blank? You know like, I think he's a little bit cluey, and I think sometimes he can… I think sometimes he can manipulate the situation a bit. But it's just really hard to know what is bad wiring and what's genuine and what's pulling our leg, you know? [P3] There would have been warnings, there would have been an explanation before we go outside for how long we are going to be out there for, what we are going to be doing, and that when I'm finished doing what I'm doing, then we will be coming inside. And what he will do is he will say yes I understand, yes I know. Yet… although he is saying the words… he's not and yet if I ask him to repeat that back to me, he will become very angry [P7]
Lying and confabulation	I know that my son does not lie ever. If you accuse him of lying, he goes absolutely ballistic [P1]
	Things that are dreams, she'll think dreams are a memory. Something that actually happened, she has told me two different things especially that she has done that I know for a fact she never has. [P4] So behavioural issues could be, you know, she has a tendency to lie a lot. And it's almost like a natural thing for her, you know. We have to then coach her into telling us the truth. And it might be just as simplest of things and so unnecessary. [P5] Yes, he does lie often. [P7] He has his moments but yeah… it's never, ever his fault. You know, if you sit down and explain to him about it all, usually he'll own up in the end but it takes you a bit to coax it out of him. [P9] When say they are outside and if they've had fight and they come in- yes. Again, he'll be working it- and I know that he has done the wrong thing, and he'll be saying, "They did this to me, and they…" and they haven't. So yeah, I suppose it is, yeah [P10] They are getting better [at not telling lies]. They are getting better, but they do [P12] (Partner) is saying that the lying, and her perception of reality is quite different from everyone else's. [P13] And, so he comes and he tells you exactly what he has done wrong and that's one thing that I've found with him but I've heard some parents say that their kids lies, but I do know that he doesn't lie.[P14]
Major theme: Adaptive Functioning (Social Skills)	
Difficulties making and keeping friends	He kind of isolates himself a bit so he… I see them more as acquaintances because his social skills are not very good. He's not the sort of kid who is very confident in walking up and talking or mixing or things like that [P1] Social skills difficulties. With the eldest one, he's not comfortable with children his own age. He is more comfortable with adults or with much younger children [P2] The other thing, he's quite popular but he doesn't really like being with people. He's actually quite happy to stay at home. If he does have

Subtheme	Exemplar quotes
	peer interaction, he is good one-on-one. So, he's better to visit people, to be out of his environment, and he's better with one kid. [P3] Her friendships change constantly, and they have mentioned that at school several times, that you know she doesn't seem to be able to keep friendships and they keep having to have these friendship conversations with her about how you treat friends and that kind of thing. [P4] She can have fun but she tends to be… because her… because she is so young-minded she likes to play younger games, or she's… I don't know. She can talk quite baby-ish if she wants to and her peers don't like that. We see that being a pattern with her, like, they are irritated by it you know "why is this doing this?". They tend to… separate from her. [P5[So he does struggle. He's got his few little friends and that's it. [P6] He just could not play along- either alongside with or do anything with this other boy. [P7] Very poor social skills. [P10] She does tend to get left behind a little bit if they are playing more complex games but they are very good at including her anyway [P11] They have big trouble making friends and keeping friends. We have been invited to zero birthday parties, zero sleep-overs. Other kids their age get to do all of this sort of us, but they don't get to do it, they don't get invited. [P12]
	I often ask "is he engaging socially in an appropriate way?", and it's becoming more obvious as the year goes on that he is quite different to the rest of the kids [P14]
Understanding social cues	An example could be say he's mucking around, or doing something, and he does something silly, I might say to him "oh you goose, why would you do that?" where another kid would laugh it off and he would go "why would you call me a goose, why don't you like me, why do you hate me, why do you call me names?" and I would go, "no sweetheart, I don't mean it in a bad way, I mean you are a goose, you are funny with what you did" and he would take that as a personal attack straight to the heart and he becomes very, very defensive, you can't talk to him about it. [P1] So, they are at that age when there is a lot of sports going on and he doesn't really understand contact sports like where the contact starts and where it ends. He can't read the other kids, say he gets tackled in football, that they aren't trying to hurt him or he'll sort of try and do the same thing back and he doesn't know when to stop. So, he finds it really hard to understand the limits of that. [P2] She can talk quite baby-ish if she wants to and her peers don't like that. We see that being a pattern with her, like, they are irritated by it you know "why is this doing this?". They tend to… separate from her. He doesn't pick up on those social cues I guess. [P6] Yeah, mostly it's… umm… emotional stuff and tones of voices is fairly big thing. So if my tone of voice changes, it might be that I'm focusing, maybe I've just had a phone call, I've got to think about what it was about and I just be writing it down. My tone of voice- he might just say something to me, and I'll say, "look, I just want to write this down, thanks, I'll come see you as soon as I've finished". He won't like that. [P7] He is very social, but it's just trying to teach him those social skills of when you are getting in someone's personal space which he doesn't

Subtheme	Exemplar quotes
	understand. But he doesn't like it if they do it to him- he gets quite upset if someone gets in his personal space. [P10] P1: But understand really the appropriateness of well maybe this boy doesn't like me being so close to him. That social cue… of I need back off a bit [P12] P1: Okay so, if anybody gets hurt in the classroom, she is the first one there giving them a hug, crying with them, a lot of boys can't cope with that, so they are like to the teacher "get her off". You know, I don't… she's too close. P2: She's got empathy. P1: Lots of empathy. P2: but doesn't know she is being… P1: But understand really the appropriateness of well maybe this boy doesn't like me being so close to him. That social cue… of I need back off a bit [P13] Whilst he is very social, he is unable to read other people's body language or emotions very well. [P14]
Indiscriminately friendly	[P13F]: Incredibly friendly. [P13M]: But once again it is inappropriate, the full on from first meeting. [P13F]: Well, she hasn't read the cues to see if she can trust this person. She's just gone all in and… It's quite scary to see, and it comes back to a vulnerability when she gets older. [P13] He would go home with anybody because he just has no awareness of danger and things like that. [P6]
	Yes, we've had zero stranger danger, it did take me a while to notice as being in out of home care, I used to think it was just because she'd had people picking her up for contact, but then as contact dropped down, she still had zero stranger danger and even now she'll just walk up to someone, sit with them, everything. [P11] Especially when she was younger. As she got older I talked a lot about stranger danger and I just said, "it's not a thing, you just don't walk up to someone because that lady said hello and mummy said hello back" that you think it's okay that you go up and cuddle them, that's just not how it works. You know, so she understands now. So, she knows now, so if someone says hello now, she'll say hello but that sort of side that I'd got out of her [P8] She's very… pretty much always happy. Very outgoing, very friendly, everyone is her friend, even though she can't keep them- everyone is her friend. And she'll invite people- oh god- over to the house all the time If some stranger walks through the door or someone that you know, and he just goes into this overdrive mode- leaping on the lounge like Tom Cruise gone crazy.
Major theme: Adaptive Functioning (Motor Skills)	
Gross motor skills	He's very… ummm… He does have a lot of co-ordination and balance. There's a lot of problems in that area. He can't balance. We are seeing an OT, and she put these blocks together for him to walk on but he struggles with the balance on it as well. [P1] So, when he came to us and we are on a few acres and we've sort of got a massive yard, and for him to be able to run around and he's wasn't even really used to outdoors. They were kept inside all the time.

Subtheme	Exemplar quotes
	He'd go to run and just topple over, and just to climb or anything, he really had no idea. [P2] I've just thought of a memory of him playing soccer, so he would have been quite young- maybe seven- and he's just a knucklehead, he'd just sort of dance around the field, I think that's because he couldn't quite control himself. [P3] He is very uncoordinated and those sorts of things. [P6] Right from the word go I did a lot of stuff with him, but even with that his balance is still a problem. He couldn't ride a bike, until he was probably about grade two, grade three until he could not have training wheels. And then, swimming he still cannot come over to breathe- to the side- without rolling all the way over. [P7] He's got poor motor skills. [P10] She has had difficulties with gross and fine motor skills. [P11] Skipping is just… I don't know if that's ever going to happen (laughter). Maybe never with the hula hoop, I just don't know. But it certainly is a real challenge for her. We took her to gymnastics, and it's just really, really hard for her. [P13] That's the, that's the amazing thing about this child. Ummm… I think… but his gross motor skills are slightly above average for his age. Which is actually very alarming. [P14]
Fine motor skills	So, his writing is still like a prep grade one-r. Right. With our eight-year-old, we've just had an assessment done by an OT, so he's three years behind in his fine motor skills, say with his handwriting.
	Well with his fine motor skills, he's a very poor writer, really poor writing. That's bad. However he's a very good drawer, and he is very good at art. Her fine motor skills, she's not very good either. He struggles a lot with hand-writing. her motor skills were behind, and that's pretty much it. I: Sure. And how about his motor skills- his hand-writing and that sort of thing? He's sort of average or below average but you know, he's getting better. [P9] At school, he does struggle at school like I said, with his fine motor skills- hand writing, all that sort of stuff. [P10] She can however hold a pencil and scissors, everything- that's really quite good. She's just lacking that strength in her hands but at least she'll be able to write. We don't have concerns about that part. [P11] I: And just keeping on the motor skills- is their hand writing generally okay? P: Yeah generally okay, it just depends on the mood though. [P12] She loves to help, but the skills just aren't there to be able to help. There is no fine motor control to be able to… say she really wants to help with the tea, well there is very limited things in what she can do because she can't actually do so many things. So what we actually end up doing is just hand over hand- "Oh you want to chop up the vegetables, alright then" so she can get the vegetables out but she wouldn't be able to do what … any other… child her age could do [P13]

Subtheme	Exemplar quotes
	He can't really use cutlery, and he tends to still want to feed himself with his fingers. He'll push food onto his spoon, and then hold the spoon in two hands to bring it up to his mouth. He can manage an open cup. Umm… but doesn't even have dagger-grip. I don't think I've ever seen children hold pens or crayons the way he does and he's just not interested in it. [P14]
Sleep concerns	As he started getting to around 4-years-old or something, the sleeping at night was shocking right up until he was diagnosed [at 7 years old], it would take me until two or three o'clock in the morning to get him to sleep. That was doing every single possible thing you know, to calming music, to no stimulation whatsoever before bed, to sitting with him with stories. He just could not sleep. And then, I would have to wake him up at 7[am] for school and he would just be, oh my god, so bad. [P1]
	I mean it's really, yeah, without the sleeping tablets, it is very, very difficult. Because what he would tend to do, and even now at times, he'll get up at night times and that when he does a lot of his thieving and stealing. [P3]
	Well, she's not too bad once she goes to sleep. But sometimes, just getting her to go to bed and she'll stay awake for hours if you don't send her. Once you send her, she sort of goes to sleep but she'll get up and do things and make a mess sometimes and stuff before she goes to bed. She'll watch movies or something. [P4]
	Very light sleep, very light sleeper. But was a very bad sleeper before, but yeah, yeah… Because we are aware of these different things as well, I mean, I used to get up three or four times a night because it was so bad. [P5]
	But the oldest one takes medication to go to sleep or he'd never stop. Ever. So he takes Catapres and he is tired by the end of the day, and he just now puts himself to bed. [P6]
	No, he has to have melatonin to get to sleep. He's a really light sleeper, so the slightest thing will wake him and he gets very angry. Generally once we are in bed as well, he will sleep through until about half past five. And we have several doors closed between us and him and even then, he will still hear us. [P7]
	He has really bad nightmares- doesn't sleep very well at all. [P10]
	No matter how much the bedtime routine doesn't change, I think she'll always fight bedtime. [P11]
	Yeah, it's challenging when you hear two children playing in the middle of the night and you look at the clock and it's two a.m., or four a.m. I've got floating shelves and I've found them jumping off the shelves on to beds, trampolining. It's dangerous! [P12]
	What we realised, what he's doing is having the equivalent of a very long nap at the beginning of the night, so when we are going to bed at about ten o'clock, it's not usual to have him sitting on the stairs, waiting as we come up the stairs. You realise that you are not going to get to sleep for about another four hours because he's up, and you can't trust that he will stay in his room- because he won't. [P14]
Cross cutting theme	
Young for age	But he's finding because his maturity is probably two or three years under the other kids in his class and he wants to hang out with girls and their maturity is sort of advanced to the boys, that now being grade

Subtheme	Exemplar quotes
	five there's real problems coming in because he doesn't understand all this girl stuff now. [P2] His behaviour is very young- very childlike. So, he doesn't act out situations like a ten-year-old should, so he is probably more like a seven-year-old, that kind of age group. [P3] But it was in kindergarten where they were starting to say that her concentration levels were poor, that you know, she was unable to sit still and all that kind of thing. That she was having trouble with maths and numbers… that was when people sort of first started noticing it. And my partner who is a teacher, like I said, had actually said to me way before that "I don't think… she's not operating at the level that she should be" [P4] she's very immature for her age, but of course that's with the FASD. [P5] I think he is just like one or two years below his peers socially and emotionally [P6] My son is not going to have a relationship with someone his own his age that is going to be positive most likely. Because other kids sense "there is something different here, and I, I don't like it and I don't want to be around it" whereas either with younger children or other children with disabilities, he is able to, he sort of tends to try to teach them a little bit I guess. [P7] I mean is still not her level for her aging, but then her aging is not a true nine-year old; her true brain capacity [P8] He acts a lot younger than his siblings [P10] We… because our daughter is only three months older… and obviously children develop at different rates but we just notices that the gap was getting bigger and bigger. And our daughter was by no means advanced, so we knew she had to falling behind a little bit [P11]
	I've got a four-year-old grandson, and they absolutely adore him and get on really well with him. But I can see that he is more… I can tell him once and he gets it, and they are starting to notice as well. They've actually said to me- "How come he gets it straight away and we don't, are we just naughty?", "No, you are not naughty, you've got a brain injury, it's not your fault". But yeah, he does seem older than them, even though he is half their age nearly. [P12] But she will just walk up and takes something off a child that she thinks would be fun to play with. Whereas you think by six you would "oh yeah, that kids is going to cry if I take that off them, I'll just (unclear) with this shiny red car, maybe I can distract them with this and take the other toy away [P13] At kindergarten, they work to the common denominator of a standard four-year old child. And he's just so far outside of that. [P14]
Inconsistency in behaviour and strategies	It's almost like a can't really win situation sometimes. If you are silly enough to think that he has recalled it [holding hands when crossing the road] this time… there is a good chance that he hasn't. And the days where he does, he gets really upset that you are [holding his hand when crossing the road]… so getting out of the car to go into Kindergarten can be, you know, a life or death experience on a daily basis. [P14] He is just very draining, very trying you know. Because, because any, you know, there is heaps of programs out there and help strategies and do this, or say this, or make this chart, use this timer, and there is a

Subtheme	Exemplar quotes
	whole of things and you can use these things to a degree but I think dealing with a child like that, you just lose the energy, you haven't got the energy to do it. And it's all very well for people to say, "oh my child responds to this kind of discipline measure, or something, but he's, he's too variable It's something that happens in the processing in the brain and there are days when it is worse than others And I remembered the teacher the year before that was saying to me, that they have days where she- her memory seems really good and she says to me it's probably not the best but we try to squeeze as much information in her as possible on those days. She said, because, I can guarantee a couple of days later she'll remember at least half of what we've told her but at least something is retaining. That was what they were trying to do, I don't think they particularly did that last year because I'm not sure if that's a good thing or bad thing when they do that. And it does sometimes get through, but other times… it's like a switch. Either she's on or she's off sort of thing. Sometimes she does understand it, sometimes she doesn't understand it. And it just depends on what headspace she's in. And where she has got one-on-one attention, she tends to get through and quite well. So we are surprised at what she can do at home and what she does at school… are very, very different you know what I mean. Sometimes, or it can be the day after. Usually by the middle of the week, if she is quite tired from big days at school, she can be erratic the next day, like back-chatty, like a meltdown, or she'll get to a stage that either they can recognise it, or she'll recognise it, and they'll get her back into a better place, where she goes into the sensory room. [P8]

Appendix B: Qualitative Study Ethics Approval (Protocol number: 0000036058)

Human ethics: Application approved

no_reply@unisa.edu.au

Fri 24/02/2017 10:36 AM

To: McDougall, Stewart Douglas - mcdsd001 <stewart.mcdougall@mymail.unisa.edu.au>; Andrea Fielder <Andrea.Fielder@unisa.edu.au>; Fiona Arney <Fiona.Arney@unisa.edu.au>;

Cc: humanethics@unisa.edu.au <humanethics@unisa.edu.au>;

Dear Applicant

Re: Ethics protocol "Enhancing screening for young children with Fetal Alcohol Spectrum Disorder: Study 1- caregiver, researcher and clinician interviews." (Application ID: 0000036058)

Thank you for submitting your ethics protocol for consideration. Your protocol has been considered by the E3 Committee Review Group.

I am pleased to advise that your protocol has been granted ethics approval and meets the requirements of the National Statement on Ethical Conduct in Human Research. You may now commence your research.

Please regard this email as formal notification of approval.

Ethics approval is always made on the basis of a number of conditions detailed at http://www.unisa.edu.au/res/forms/docs/humanresearchethics_conditions.doc; it is important that you are familiar with, and abide by, these conditions. It is also essential that you conduct all research according to UniSA guidelines, which can be found at http://www.unisa.edu.au/res/ethics/default.asp

Please note, if your project is a clinical trial you are required to register it in a publicly accessible trials registry prior to enrolment of the first participant (e.g. Australian New Zealand Clinical Trials Registry http://www.anzctr.org.au/) as a condition of ethics approval.

Best wishes for your research.

Executive Officer
UniSA's Human Research Ethics Committee
CRICOS provider number 00121B

This is an automated email and cannot be replied to. Please direct your query to humanethics@unisa.edu.au.

Appendix C: Qualitative Study Recruitment Materials

UniSA | Australian Centre for Child Protection

Improving screening for young children with fetal alcohol spectrum disorder.

Are you currently caring for a young child who has been diagnosed with a fetal alcohol spectrum disorder?

We are seeking parents and carers currently raising a young child who has been diagnosed with a FASD to take part in the first stage of a research project aiming to enhance the screening of young children suspected of having FASD.

We would like to invite you to participate in a telephone interview to discuss the challenges experienced by children with a FASD that you are caring for, or have cared for.

You will be reimbursed for your time with a $20 Cole Myer gift-card.

If you would like to participate, please contact Stewart McDougall, or leave a message with your contact details and we will contact you

Telephone or SMS	(08) 8302 2983 *or* [redacted]
Email	Stewart.mcdougall@unisa.edu.au
Surveymonkey link	Provide contact details and we will contact you https://www.surveymonkey.com/r/7QLSPH7

We understand the challenges associated with obtaining a diagnosis may mean that you strongly believe that a child has FASD but has not received a diagnosis yet. If this describes your situation, please contact the research to discuss your potential participation in the research project.

This project has been approved by the University of South Australia's Human Research Ethics Committee. In accordance with ethics procedures, data will only be accessible by the researchers and confidential data will be securely stored by UniSA for 5-years prior to being destroyed.

If you have any ethical concerns about the project or questions about your rights as a participant, please contact Vicki Allen, the Executive Officer of the Ethics Committee, Tel: +61 8 8302 3118; Email: Vicki.allen@unisa.edu.au

Appendix D: Qualitative Study Interview guides

Caregiver interview guide

Prior to interview commencing

- Explain the purpose of research
- Re-confirm that the participant is interested and willing to participate
- Read information and consent sheet, answer any question. Indicate participant's consent.
- Start recorders after consent, and let participant know that the recorders are running

Stage 1: Introduction and demographic details

- What is your relationship to your child/ren with diagnosed with or suspected of having
- FASD?
- What gender is/are these child/ren
- How old are these children?
- How old were children when the diagnosis was conferred?
- What diagnosis did your child receive?

Stage 2: Body of the interview

- What are some of the cognitive and behavioural difficulties that your child experiences?
- How are their relationships with other children, peers, or siblings?
- How are they performing in school?
- When did behaviours being to emerge, and what led you suspect that this child may have
- FASD?
- What areas of strength do they display?
- Is there anything else you would like to add?

Stage 3: Closing off the interview

- Make arrangements for sending transcript for additional comments, and gift card

Clinician Discussion Guide

Interview background

- - Undertaking PhD research looking at improving the screening for FASD in young
- Children. - Interviews with clinicians as part of a broader program of research following a line of research looking at both behavioural measures and cognitive tests to identify children
- with FASD
- - Interested in clinicians views on what would be the most useful domains (such as those highlighted in the Australian diagnostic criteria) to include in a screening tool, understanding the diagnostic process and where screening may best fit.

Interview questions

- What behaviours are children most commonly referred to assessment for?
- What behaviours are the biggest concern for parents and carers?
- In your view, of the domains highlighted in the diagnostic guidelines, which, if any, would be most useful or informative to capture in a screening tool?
- What tools do you currently use in the assessment and diagnosis of FASD?
- Any additional information that a screening tool could also aim to capture? (e.g.,
- developmental milestones,
- Questions to capture the development of children with FASD; does the presentation of
- FASD change with age? Gender differences?

Demographics

- Role
- Length of time working with kids with FASD
- Number of patients that you see with FASD a year
- % of patients seen per year

Appendix E: Content Validation Questionnaire

FASD Screening tool content validation

1. FASD Screening tool

Thank for your interest in this study.

The purpose of this study is to seek feedback from caregivers and clinicians of children with FASD on the initial findings of a program of research investigating ways to improve screening for FASD. Further, feedback is being sought on the proposed content of a screening tool for children at risk of having FASD.

There are two parts to this research project.

Firstly, you will be presented with the key findings from the initial interview study that conducted interviews with caregivers and clinicians to investigate the behavioural and cognitive difficulties faced by children with FASD. You will be asked to provide any comments on the findings in a text box.

Secondly, you will be presented with a series of 98 items and asked to read each item carefully before rating on a 4-point scale and provide any comments on whether or not the item is appropriate, and whether you think the item is clear and concise, or could benefit from being re-written, or even omitted.

It is anticipated the this will take between 15 and 30 minutes.

If you wish to participate, please indicate below and proceed to the survey, otherwise click "No" and close this window.

Your participation in this research will be voluntary and anonymous, and participation in this research will in no way impact upon the services or supports that you may be accessing through any organisations. Participants are free to discontinue the survey at any time. An individual's decision to not participate or to discontinue will not result in any consequences now or in the future. While completing this survey participants may experience feelings of mild distress or discomfort; however, we do not foresee the possible risks to be greater than those encountered during everyday life.

All records containing personal information will remain confidential and no information which could lead to identification of any individual will be released, unless required by law.

If you have any questions regarding this study, please contact Stewart McDougall Tel: 08 8302 2983; Email: stewart.mcdougall@unisa.edu.au, Professor Fiona Arney Tel: (08) 8302 2918; E: fiona.arney@unisa.edu.au, Dr. Andrea Gordon Tel : (08) 8302 1764, Email: andrea.gordon@unisa.edu.au, or Dr. Amy Finlay-Jones Tel: (08) 9489 7609, Email: amy.finlay-jones@telethonkids.org.au.

A summary or a copy of the research report will be made available to you if you wish once the study has been completed.

This project has been approved by the University of South Australia's Human Research Ethics Committee. In accordance with ethics procedures, data will only be accessible by the researchers and confidential data will be securely stored by UniSA for five years prior to being destroyed.

If you have any ethical concerns about the project or questions about your rights as a participant, please contact Vicki Allen, the Executive Officer of the Ethics Committee, Tel: +61 8 8302 3118; Email: Vicki.allen@unisa.edu.au. Participants or third parties who wish to lodge a complaint about either the study or the way it is being conducted should contact the Executive Officer of UniSA HREC in the first instance, email: humanethics@unisa.edu.au or tel: 8302 3118

* 1. Do you wish to particpate in this study?

- ◯ Yes
- ◯ No

FASD Screening tool content validation

2.

2. Are you the parent or carer of a child with a FASD, or a clinician working with children with FASD?

- ◯ Parent or carer
- ◯ Clinician

3. Please select the option which best describes your gender?

- ◯ Male
- ◯ Female
- ◯ Other (please specify)
- ◯ Prefer not to answer

4. Please enter your age in years

5. For parents and carers only, please indicate your relationship to the child/ren you care for

- ◯ Biological parent
- ◯ Adoptive parent
- ◯ Foster parent
- ◯ Step-parent
- ◯ Grandparent, aunt, uncle, or other relative
- ◯ Other (please specify)

6. For parents and carers,

How many children with FASD are you raising?

How old is this child/these children?

What gender is this child/these children?

What diagnosis, if known, did they receive?

7. For clinicians working with children living with FASD, please indicate your profession

- ◯ Pediatrician
- ◯ Occupational therapist
- ◯ Psychologist or neuropsychology
- ◯ General practitioner
- ◯ Speech pathologist
- ◯ Other (please specify)

8. How long have you worked with, or been raising a child/children with FASD?

FASD Screening tool content validation

3. Findings from stage one of the research

9. The image above shows a mind-map of the key themes (*green)* and sub-themes (*yellow*) identified relating to the common cognitive and behavioural difficulties experienced by children with FASD and their caregivers in the preceding phase of the research. Please consider these themes and provide any comments you may have. For example if you feel that we have missed anything important, please provide this feedback in the comment box below.

If you have no comments, please leave the box blank and move on to the next question.

FASD Screening tool content validation

4. Screening tool validation

Children with FASD are often missed as having nothing wrong, or mis-diagnosed. A screening tool which can accurately identify children at risk of FASD may aide caregivers, teachers, and health professionals to raise their concerns about the child's behaviour, make appropriate referrals for further assessment, and to provide directions for interventions whilst families are waiting for assessments to be conducted. Currently there are no valid screening tools for FASD.

We are seeking your feedback on the proposed content for a new tool intended to assist parents and carers to identify if their children may be at risk of having a FASD.

The tool will consist of a series of behavioural questions intended to measure attention, concentration, impulsivity and hyperactivity, and social relationships. The questions have been developed through interviews with caregivers and clinicians and through a review of the scientific literature about the challenges experienced by children with FASD.

The tool is intended to be completed by caregivers who have concerns about their child's behaviour in the early years of primary school.

As this tool is intended to identify children at risk of FASD, please consider your child's behaviour at the time they were diagnosed.

Instructions

Please read each item carefully. After you have read the item, consider whether the item reflects your experience raising a child living with FASD, and then rate the item on the four-point scale.

1= Not relevant at all: this item does not reflect your experience, and is not a behaviour that your child has displayed.
2= Somewhat relevant: this item does not really reflect *your* experience, but might reflect the experience of others.
3= Quite Relevant but needs rewording: this item reflects your experiences, but could be improved
4= Highly relevant: this item strongly reflects your experience, and should be retained in a screening tool.

If the item is relevant but needs re-wording, or you wish to make comment on the item, please use the comment box below each item.

A comment box is provided at the end of the questionnaire to provide any additional comments, and to suggest any other items which may be relevant.

If you have any questions or difficulties, please contact Stewart McDougall (08 8302 2983 or 0433 670 749), and stewart.mcdougall@unisa.edu.au.

FASD Screening tool content validation

5. Hyperactivity and impulsivity

10. Please rate the following items on how relevant you believe the item is to the behaviour of your child/ren living with FASD.

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Talks excessively	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Runs about or climbs excessively in situations in which it is inappropriate	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Leaves seat in classroom or in other situations in which remaining seated is expected	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Interrupts or intrudes on others (e.g., butts into conversations or games)	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty playing or engaging in leisure activities quietly	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty awaiting turn	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Is often "on the go" or acts as if "driven by a motor"	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty stopping an activity immediately upon being told to do so	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty refraining from smiling or laughing in situations where it is inappropriate	○	○	○	○

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Please fill out the box below if you wish to make a comment on this item				
Has difficulty holding back his/her activity despite being told to do so	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Does things without first thinking about what could happen	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Acts in a wilder way compared to other children when in a group (e.g., at a birthday party, during a group activity or on a school excursion)	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Requires constant supervision	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Does risky things	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

FASD Screening tool content validation

6. Memory, understanding consequences, and academic ability

11. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture memory, understanding consequences and academic ability

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Appears not to have heard what you say and appears to ignore you	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Struggles with simple maths concepts (e.g., counting, arithmetic)	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Struggles with complex maths problems (e.g., multiplication, word problems)	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Struggles with literacy	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulties comprehending written information	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Is assigned schoolwork at a grade level below their current grade	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Does not see the whole picture	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Difficulties with abstract thinking	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has trouble remembering to do things like school projects or chores	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Often needs things repeated a number of times before they start to remember	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Needs repeated exposure to learning things properly	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Forgets things from one day to the next (for example, forgets school work that they knew the night before)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Struggles to recall recent information such as what just happened in a story, or what was just said to them	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Struggles to remember activities that occurred during the day (e.g., what they did in the morning, what they ate)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Often loses their lunch box, school work sheets, permission slips	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Has difficulties anticipating or understanding the consequences of their actions	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Does not display remorse	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Makes the same mistakes repeatedly	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Behaviour does not improve despite consistent consequences	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Doesn’t attempt to hide evidence of misbehavior (e.g., tells you when they have done something they should know is wrong)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Inflexible behaviour, for example gets stuck on an idea or object	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

FASD Screening tool content validation

7. Emotional regulation

12. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture emotional regulation.

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has outbursts for little reason	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has angry or tearful outbursts are intense but end suddenly	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Exhibits wide mood swings	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has frequent meltdowns	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Can’t be rushed between activities	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has tantrums that last longer than would be expected from a child of that chronological age	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty handling exciting events	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Is agitated if not getting their way	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Resists changes of routine	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Is overwhelmed in new situations or places	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Is more difficult to manage in public than at home	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

FASD Screening tool content validation

8. Argumentative, or aggressive behaviour

13. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture argumentative, or aggressive behaviour.

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Is aggressive towards siblings, peers or adults (e.g., parents, teachers)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Destroys own property, or family's property	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Often argues with adults	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Often quarrelsome or bossy with peers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

FASD Screening tool content validation

9. Attention and concentration

14. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture attention and concentration.

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has trouble keeping track of what he/she is doing	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Is easily distracted by other things around them (e.g., other kids, things going on around them, background noise)	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Needs help from an adult to stay on task	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has trouble remembering a list of tasks	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has trouble completing tasks that have more than one step.	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has trouble remembering things, even for a few minutes	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

FASD Screening tool content validation

10. Social skills and peer interaction

15. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture social skills and interactions will peers.

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has an air of competence	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Appears more capable than he or she really is	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Acts on own needs or doesn't consider the needs of others	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has superficial friendships; is a social butterfly	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulty making or keeping friends	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Engages appropriately in group play	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Is not aware of his or her actions on others	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Doesn't understand personal space	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Behaves in a way towards other children which they know annoys themselves	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Does not understand social cues (e.g., facial expressions, tone of voice, body language)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Indiscriminately friendly towards strangers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Easily led by peers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Acts emotionally and socially younger than peers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Acts young for age	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Prefers the company of younger peers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

Able to engage more readily with younger peers, compared to same-age peers	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

FASD Screening tool content validation

11. Language and communication skills

16. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture language and communication skills

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Can't take a hint	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Chatty, but with little content	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Uses simpler language than peers	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Conversations with your child are often difficult for a listener to follow	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Uses poor grammar in spoken sentences	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Has difficulties understanding jokes	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Replaces fact with fantasy	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Lies when it would be easy to tell the truth	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				
Follows along with what is said/suggested, even if not true	○	○	○	○

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Please fill out the box below if you wish to make a comment on this item				
Appears sincere when caught out in a lie	○	○	○	○
Please fill out the box below if you wish to make a comment on this item				

FASD Screening tool content validation

12. Daily living skills, sleep, and motor skills

17. Please rate the following items on how relevant you believe the item is to the behaviour of children with an FASD. These items are intended to capture daily living skills, sleep, and motor skills

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has trouble learning and using the concept of time	○	○	○	○
Difficulty understanding and applying concepts such as now, or later, or days of the week	○	○	○	○
Resists going to bed at night	○	○	○	○
Is a light sleeper	○	○	○	○
Does not dress appropriately for the weather	○	○	○	○
Decreased awareness of pain or temperature	○	○	○	○
Poor handwriting	○	○	○	○
Difficulty handling, assembling, and manipulating small objects	○	○	○	○
Does not hold a crayon/pen/pencil properly	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has difficulty throwing and catching a ball	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has difficulty running fast	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has problems with balance (e.g., struggles to stand on one leg)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

	Not relevant	Somewhat relevant	Quite relevant, but needs to be reworded	Highly relevant
Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	○	○	○	○

Please fill out the box below if you wish to make a comment on this item

FASD Screening tool content validation

13. Summary page

18. If you have any additional comments, or would like to suggest any items which we may have missed, please use the comment box below.

Thank you for participating as a research participant in the present study concerning whether a newly developed tool can be used to identify children at-risk of being diagnosed with fetal alcohol spectrum disorder. Your participation is greatly appreciated.
If you have any questions regarding

If you have any questions, please contact Stewart McDougall at stewart.mcdougall@unisa.au or on (08) 8302 2983, or 0433 670 749.

Once again, thank you for your participation.

Appendix F: Preliminary Reliability and Validity Study Ethics Approval (Protocol Number: 200987)

McDougall, Stewart Douglas - mcdsd001

From: no_reply@unisa.edu.au
Sent: Friday, 12 October 2018 2:24 PM
To: McDougall, Stewart Douglas - mcdsd001; Fiona Arney
Cc: Human Ethics
Subject: Human ethics: Application approved

Dear Applicant

Re: Ethics protocol "Validation of a new FASD screening tool" (Application ID: 200987)

Thank you for submitting your ethics protocol for consideration. Your protocol has been considered by the E3 Committee Review Group.

I am pleased to advise that your protocol has been granted ethics approval and meets the requirements of the National Statement on Ethical Conduct in Human Research. You may now commence your research.

Please regard this email as formal notification of approval.

Ethics approval is always made on the basis of a number of conditions detailed at http://www.unisa.edu.au/res/forms/docs/humanresearchethics_conditions.doc; it is important that you are familiar with, and abide by, these conditions. It is also essential that you conduct all research according to UniSA guidelines, which can be found at http://www.unisa.edu.au/res/ethics/default.asp

Please note, if your project is a clinical trial you are required to register it in a publicly accessible trials registry prior to enrolment of the first participant (e.g. Australian New Zealand Clinical Trials Registry http://www.anzctr.org.au/) as a condition of ethics approval.

Best wishes for your research.

Executive Officer
UniSA's Human Research Ethics Committee
CRICOS provider number 00121B

This is an automated email and cannot be replied to. Please direct your query to humanethics@unisa.edu.au.

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Appendix G: Preliminary Reliability and Validity Study Recruitment Materials

UniSA | Australian Centre for Child Protection

Improving screening for young children with fetal alcohol spectrum disorder.

Are you currently caring for a young child who has been diagnosed with fetal alcohol spectrum disorder? Or is suspected of living with fetal alcohol spectrum disorder?

We are seeking parents and carers raising children between the ages of 4 and 12 years old to take part in a research project aiming to enhance the screening of young children suspected of having FASD.

Parents and carers of children who are not living with Fetal Alcohol Spectrum Disorder are strongly encouraged to participate as we need your input to serve as a comparison group.

The link below will take you to the study webpage containing more information about the study. If you choose to take part you will be asked to follow a link to the survey, and then complete a number of questions about yourself and your children before completing a questionnaire about your child's behaviour. The survey is anticipated to take approximately 40 minutes to complete, and upon completion all participants will have the opportunity to enter a raffle to win one of eight $50 gift cards.

(INSERT STUDY WEBPAGE URL)

If you would like any further information, please contact Stewart McDougall, or leave a message with your contact details and we will contact you

Telephone or SMS [redacted]

Email [redacted]

This project has been approved by the University of South Australia's Human Research Ethics Committee. In accordance with ethics procedures, data will only be accessible by the researchers and confidential data will be securely stored by UniSA for 5-years prior to being destroyed.

If you have any ethical concerns about the project or questions about your rights as a participant, please contact Vicki Allen, the Executive Officer of the Ethics Committee, Tel: +61 8 8302 3118; Email: Vicki.allen@unisa.edu.au

Appendix H: Preliminary Reliability and Validity Questionnaire

FASD screening tool pilot study

Participant information and consent sheet

Why is the research being conducted?

Children living with Fetal Alcohol Spectrum Disorder (FASD) experience significant cognitive and behavioural difficulties in their lives that may go unrecognised. As a result, children and families may not receive the help they need to reach their full potential. Early and accurate identification of children at-risk of having FASD is vital to ensuring children and families receive the help they need.

This project aims to test if a newly-developed screening tool can accurately identify children at-risk of having FASD from their peers who are not at-risk.

Australian and New Zealand parents or carers, currently caring for a child with FASD or is suspected to have FASD between the ages of 4 and 12 are welcome to participate. Parents and carers of children who do not have FASD are strongly encouraged to participate as a comparison group.

All participants will have the opportunity to enter a raffle to win one of eight $50 vouchers upon completing the survey.

What will you be asked to do?

You will be asked to complete an online survey focussing on the behaviour, social skills, and cognitive abilities of your child.

The survey also includes short demographic questions where you will be asked about your age, education, lifestyle during pregnancy, and whether your child/ren have any health concerns. Some questions are of a sensitive nature and participants may experience some discomfort in answering these questions. If you experience discomfort or distress during the survey, please exit the survey and consult the list of available supports provided on the study website (http://www.unisa.edu.au/Research/Australian-Centre-for-Child-Protection/Research/Testing-a-new-screening-tool-for-children-living-with-FASD/). Please contact the services, or discuss your concerns with a health professional.

Caregivers of children with FASD will also be asked about the diagnostic process for FASD.

Participation will take approximately 40 minutes. At any point you may close the survey and come back to it later without losing any information, provided you use the same computer or mobile device.

You will be invited to opt-in for a follow-up study in two to four weeks where you will receive a link to another questionnaire which will repeat certain sections of this questionnaire. It is anticipated that the follow up session will take between 10 and 20 minutes to complete. Participation in this and the follow up study is voluntary. Should you opt-in to complete the follow up study, you will be asked to provide an email address so that a unique link may be provided to you. If you do opt-in, a unique code will be applied to both your initial questionnaire answers and your answers to the follow up session to allow the research team to compare your answers across time. All identifying information will be deleted at the earliest possible moment to ensure the confidentiality of your data.

Your Participation is voluntary

Your participation is voluntary, and you may withdraw from the study at any time prior to submitting the online survey. Should you wish to withdraw from the survey, please first click the “exit survey” button at the bottom of each page rather than the “close window” button. The exit survey button will take you to an end-of-survey page which contains contact details for the researchers and information about supports available to you, should you feel any distress.

No individual identifying information will be recorded for the survey and your participation will be completely anonymous unless you opt in for the follow-up study or unless required by law. The responses you provide will remain confidential and cannot be identified in the reporting of the research. The results of the study may be published, however, no information that could lead to the identification of any participants will be published. The researchers will take every care to remove responses from any identifying material as early as possible. Likewise, individual's responses will be kept confidential by the researcher and not be identified in the reporting of the research. However, the researcher cannot guarantee the confidentiality or anonymity of material transferred by email or the internet.

Further, as Survey Monkey is being used to collect data, data collected will be held offshore on the Survey Monkey's servers during the data collection phase of the project. Data will be downloaded from surveymonkey and held on a password protected computer at the Australian Centre for Child Protection (ACCP) for a period of five years after the completion of the project.

What are the expected benefits of the research?

While there are no immediate benefits to participants, the findings from this research will be used to help us learn more about how we can be better identify children who may be at-risk of having Fetal Alcohol Spectrum Disoder.

Risks to you

It is possible that some participants may experience some distress or emotional discomfort while completing this survey. A list of counselling and help-lines are provided on the study website. You will be directed back to the study webpage when you either use the "exit survey" button, or complete the survey. Please ensure that you use the "exit survey" button to leave the survey, otherwise you may lose this information. We encourage all participants to download a copy of the services available for your records.

Feedback to you

A summary of the research findings will be available for participants online at (ACCP website) and distributed through social media networks in February 2019. If you would like to receive a summary of results directly, please contact Stewart McDougall using the contact details below.

Questions/ Further Information

A copy of this information is available to download from the study website, should you wish to have a copy for your records.

If you have any questions regarding this study, please contact

Stewart McDougall, PhD Scholar.

Tel: +61 8 8302 2983; Email: stewart.mcdougall@mymail.unisa.edu.au

Alternatively, you may contact the project supervisors; Prof. Fiona Arney: Fiona.arney@unisa.edu.au; Dr. Andrea Gordon andrea.gordon@unisa.edu.au; Dr. Amy Finlay-Jones amy.finlay-jones@telethonkids.org.au

The Ethical Conduct of this Research

This project has been approved by the University of South Australia's Human Research Ethics Committee. In accordance with ethics procedures, anonymous electronic data will be securely stored by UniSA for five years prior to being destroyed.
If you have any ethical concerns about the project or questions about your rights as a participant, please contact Vicki Allen, the Executive Officer of the Ethics Committee, Tel: +61 8 8302 3118; Email: vicki.allen@unisa.edu.au

Participants or third parties who wish to lodge a complaint about either the study or the way it is being conducted should contact the Executive Officer of UniSA HREC in the first instance, email: humanethics@unisa.edu.au or tel: 8302 3118

* 1. **I understand what involvement in this research comprises and I consent to participate. I understand my participation is voluntary and I may withdraw from the study at any time prior to submitting the online survey.**

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Initial Screening: Australian residency

Due to the requirements of the approvals of our ethics body, we need to confirm that your a resident of Australia or New Zealand.

* 2. In what country do you currently reside?

- ◯ Australia
- ◯ New Zealand
- ◯ Other (please specify)

FASD screening tool pilot study

Initial screening: Participant age

* 3. Are you over the age of 18?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Initial screening: FASD diagnosis

* 4. Does one or more of the children you are raising have, or is suspected of having Fetal Alcohol Spectrum Disorder (FASD)?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Parent and caregiver demographics

The following questions will ask for some demographic details about yourself and your children.

5. Please enter your age in years

6. Please select the option which best describes your gender

- ◯ Male
- ◯ Female
- ◯ Prefer not to answer
- ◯ Other (please specify)

7. Please select which state you live in

8. Please select which region you live in

9. Please select the option which best describes your level of education

- ◯ Less than year 8
- ◯ Less than year 12
- ◯ Year 12, or equivalent (HSC/Leaving cert)
- ◯ Vocational qualification (e.g., Cert 1-4)
- ◯ Diploma, or associate degree
- ◯ Undergraduate qualification
- ◯ Post graduate qualification (Graduate certificate or diploma, Master's degree or Doctorate).

10. Please select the option which best describes your level of education

- ◯ Less than year 9
- ◯ Less than year 13
- ◯ Year 13, or equivalent
- ◯ Vocational qualification (e.g., Cert 1-4)
- ◯ Diploma, or associate degree
- ◯ Undergraduate qualification
- ◯ Post graduate qualification (Graduate certificate or diploma, Master's degree or Doctorate).

11. Please select the option which best describes your employment status

- ◯ Employed full time
- ◯ Employed part-time
- ◯ Carers pension
- ◯ Retired
- ◯ Studying
- ◯ Unemployed

12. Are you of Aboriginal, Torres Strait Islander, or Islander origin?

Please check all that apply.

- ☐ Yes, Aboriginal
- ☐ Yes, Torres Strait Islander
- ☐ Yes, Islander
- ☐ No

13. Which ethnic group do you belong to?

- ☐ New Zealand European
- ☐ Maori
- ☐ Samoan
- ☐ Cook Island Maori
- ☐ Tongan
- ☐ Niuean
- ☐ Chinese
- ☐ Indian

14. How many children are you currently raising?

15. How many children with FASD are you raising?

FASD screening tool pilot study

Child demographics

If you have more than one child between the ages of 4 and 12 who is living with FASD, please answer the following questions based on the child whose birthday is coming up next.

If you have more than one child between the ages of 4 and 12, please answer the following questions based on the child whose birthday is coming up next.

16. What is this child's name?

Your response to this question will be used to personalise the remaining questions, and to help you keep one child in mind. This information will be permanently deleted when the responses are downloaded from the Survery Monkey servers.

* 17. How old is this child?

18. Please select the option which best describes your child's gender

19. When did you first suspect that this child was living with FASD?

20. What first lead you to suspect that this child was living with FASD?

21. When was the diagnosis conferred/confirmed?

22. What diagnosis did your child receive?

- [ ] FASD, with sentinel facial features, or Fetal Alcohol Syndrome
- [ ] FASD, without sentinel facial features
- [ ] Unsure
- [ ] Other, please specify if known (E.g., Static Encephalopathy: Alcohol Exposed, Fetal Alcohol Syndrome, Partial Fetal Alcohol Syndrome, Neurodevelopmental Disorder: Alcohol Exposed)

23. How was this diagnosis conferred? (Single clinician, multi-disciplinary team)

24. Is this child of Aboriginal, Torres Strait Islander, or Islander origin?
Please tick all that apply

- [ ] Yes, Aboriginal
- [ ] Yes, Torres Strait Islander
- [ ] Yes, Islander
- [ ] No

25. Which ethnic group does this child belong to?

- [ ] New Zealand European
- [ ] Maori
- [ ] Samoan
- [ ] Cook Island Maori
- [ ] Tongan
- [ ] Niuean
- [ ] Chinese
- [ ] Indian

26. Which of the following best describes your relationship to this child?

- ◯ Biological parent
- ◯ Step parent
- ◯ Foster parent
- ◯ Other (please specify)
- ◯ Grandparent, aunt or uncle, or other relative
- ◯ Adoptive parent

27. For carers other than biological parents, how long has this child been in your care?

28. Does this child have any serious health conditions, in addition to FASD?
Please list all that apply

29. Does this child have any serious health conditions? (please list all)

FASD screening tool pilot study

Lifestyle during pregnancy: Carers other than biological parent supplementary question

30. The next questions will ask you about lifestyle during pregnancy. In a previous question you indicated that you were not the biological parent of this child.

When answering the following questions please base your responses on your knowledge of the birth mothers lifestyle during pregnancy.

What source, or sources, of information about the mothers life style during pregnancy are you drawing on? (Check all that apply)

- [ ] Personal relationship with the birth mother during the pregnancy
- [ ] Court, or medical documents
- [ ] Conversations with the birth mother after pregnancy
- [ ] Other (please specify)

FASD screening tool pilot study

Lifestyle during pregnancy questionnaire

The next questions relate to you, or your partners, pregnancy, and your lifestyle during pregnancy. There are no right or wrong answers we simply would like to gather information about your pregnancy. If you can be as open and honest as possible this would greatly help our research.

Fathers, please answer the questions to the best of your knowledge.

Please tick the response which most accurately reflects your experience during pregnancy.

31. What was the birth weight of this child?

32. At what gestational age was this child born?

FASD screening tool pilot study

Lifestyle during pregnancy

33. Did you take any nutritional supplements during this pregnancy?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Lifestyle during pregnancy

34. If yes, please specify which supplements you took

FASD screening tool pilot study

Lifestyle during pregnancy

35. Did you smoke cigarettes or tobacco at all during this pregnancy?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Lifestyle during pregnancy

36. If yes, during which trimesters did you smoke?
Check all that apply

- [ ] First
- [ ] Second
- [ ] Third

37. What was the average number of daily cigarettes smoked in pregnancy?

38. Did you cut down, or quit smoking during this pregnancy?

- ◯ Yes
- ◯ No

39. If you cut down, or quit smoking, during this pregnancy at approximately which week of pregnancy did this occur?

FASD screening tool pilot study

Lifestyle during pregnancy

40. Did you consume any alcohol during this pregnancy?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Lifestyle during pregnancy

41. If yes, during which trimesters was alcohol consumed?
Please check all that apply

- [ ] First
- [ ] Second
- [ ] Third

42. How often did you have a drink during this pregnancy?

- ◯ Daily or almost daily
- ◯ 2 or 3 times a week
- ◯ Once a week
- ◯ Once every 2 weeks
- ◯ Once a month
- ◯ Less than once a month
- ◯ Never

43. How many drinks did you have on a typical day when drinking?

- ◯ 10 or more
- ◯ 7–9
- ◯ 5–6
- ◯ 3–4
- ◯ 1–2
- ◯ Unknown

44. How often did you have more than 4 drinks on one occasion?

- ◯ Daily or almost daily
- ◯ 2 or 3 times a week
- ◯ Once a week
- ◯ Once every 2 weeks
- ◯ Once a month
- ◯ Less than once a month
- ◯ Never

45. Did you ever drink more, for example at a special occasion or during difficult times?

- ◯ Yes
- ◯ No

FASD screening tool pilot study

Proposed FASD screener

The next part of the survey consists of 6 questionnaires about your child's behaviour, feelings, and cognitive abilities. The first of these, is the new screening tool and is 69 items long.

As you answer these questions, please keep {{ Q16 }} in mind.

After this you will complete 5 shorter questionnaires, with between 9 and 24 items. We realise that there is some overlap between these questionnaires and you may find yourself answering similar questions throughout the questionnaires. This type of repetition is unavoidable when trying to develop new screening tools and we ask that you please complete all questions.

You may exit the survey at any point, as long as you follow the link on the same computer or device, your responses will be saved.

46. Please rate each item on how true of your child the following items are. Items are scored on a 4-point scale: 1) Never true, 2) Sometimes true, 3) Often true, 4) Very often true.

When answering the questions, please keep in mind your childs behaviour over the the past 6 months.

Compared to other children their age, my child…

	Never true	Sometimes true	Often true	Very often true
Talks much more than other children	○	○	○	○
Has difficulty playing or engaging in leisure activities quietly	○	○	○	○
Has more difficulty waiting for their turn than other children	○	○	○	○
Is often "on the go" or acts as if "driven by a motor"	○	○	○	○
Finds it difficult to stop doing something when asked to	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Appears to act without thinking	○	○	○	○
Acts in a more hyperactive or excited way	○	○	○	○
Requires constant/or high levels of supervision	○	○	○	○
Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	○	○	○	○
Behaviour is often inflexible, (e.g., gets “stuck” on an idea or object)	○	○	○	○
Runs about or climbs excessively when it is inappropriate	○	○	○	○
Interrupts or intrudes on others (e.g., butts into conversations or games)	○	○	○	○
Appears to ignore people, or not listen to what is said	○	○	○	○
Struggles with simple maths concepts (e.g., counting, arithmetic)	○	○	○	○
Struggles with complex maths concepts (e.g., multiplication, word problems)	○	○	○	○
Struggles with literacy (e.g., reading comprehension, spelling, writing)	○	○	○	○
Has difficulty understanding written information	○	○	○	○
Has difficulty understanding verbal information	○	○	○	○
Is assigned schoolwork at levels below their current year	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	○	○	○	○
Has more trouble remembering to do school projects or chores	○	○	○	○
Often needs information repeated a number of times before they start to learn and remember it	○	○	○	○
Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)	○	○	○	○
Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	○	○	○	○
Often loses their belongings (e.g., toys, lunch box, school work)	○	○	○	○

47. Please rate each item on how true of your child the following items are. Items are scored on a 4-point scale: 1) Never true, 2) Sometimes true, 3) Often true, 4) Very often true.

When answering the questions, please keep in mind your child's behaviour over the past 6 months.

Compared to other children their age, my child…

	Never true	Sometimes true	Often true	Very often true
Takes things which do not belong to them	○	○	○	○
Has difficulty anticipating the consequences of their actions	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Repeats mistakes without seeming to realise or learn from previous attempts	○	○	○	○
Does not appear to care about negative consequences to their behaviour	○	○	○	○
Has outbursts for little apparent reason	○	○	○	○
Has frequent meltdowns	○	○	○	○
Exhibits wide mood swings	○	○	○	○
Has emotional outbursts that end suddenly	○	○	○	○
Needs a lot of time to transition between activities	○	○	○	○
Is easily over-whelmed, or over-stimulated	○	○	○	○
Becomes agitated if they aren't getting their way	○	○	○	○
Has difficulty adapting to changes of routines	○	○	○	○
Destroys belongings, either their own or others	○	○	○	○
Is easily distracted by other things around them	○	○	○	○
Needs more help from an adult to stay on task	○	○	○	○
Needs tasks broken down step by step	○	○	○	○
Has trouble making friends with similar age peers	○	○	○	○
Has trouble keeping, or maintaining, friendships	○	○	○	○
Appears unaware of the impact of his or her actions on others	○	○	○	○
Does not appear to understand personal space	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Is easily led by peers	○	○	○	○
Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	○	○	○	○
Engages more readily with younger peers	○	○	○	○

48. Please rate each item on how true of your child the following items are. Items are scored on a 4-point scale: 1) Never true, 2) Sometimes true, 3) Often true, 4) Very often true.

When answering the questions, please keep in mind your child's behaviour over the past 6 months.

Compared to other children their age, my child…

	Never true	Sometimes true	Often true	Very often true
Doesn’t pick up on subtle social cues, such as body language, tone of voice, and facial expressions	○	○	○	○
Talkative, but with little depth or coherency	○	○	○	○
Unclear pronunciation of sounds/words	○	○	○	○
Restarts sentences or words multiple times, as though they can’t get the word out	○	○	○	○
Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	○	○	○	○
Takes longer than other children to process information and respond	○	○	○	○
Uses simpler words, and has less variety in words (smaller vocabulary)	○	○	○	○
Uses simpler sentences than peers	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	○	○	○	○
Appears to replace fact with fantasy	○	○	○	○
Follows along with what is said/suggested, even if it is not true	○	○	○	○
Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	○	○	○	○
Has difficulty understanding and applying the concept of time	○	○	○	○
Has difficulty understanding and applying concepts such as now, later, before, or the days of the week	○	○	○	○
Does not dress appropriately for the weather	○	○	○	○
Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)	○	○	○	○
Has problems with balance (e.g., struggles to stand on one leg)	○	○	○	○
Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	○	○	○	○
Appears to zone out or look lost in thought	○	○	○	○
Appears to have difficulty applying learning across different settings	○	○	○	○

	Never true	Sometimes true	Often true	Very often true
Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	○	○	○	○
History of recurrent ear infections or hearing difficulties	○	○	○	○

FASD screening tool pilot study

Strengths and Difficulties Questionnaire

49. **For each item, please click the box for Not True, Somewhat True or Certainly True.**

It would help us if you answered all items as best you can even if you are not absolutely certain.

Please give your answers on the basis of the child's behaviour over the last six months or this school year.

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	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	○	○	○
Restless, overactive, cannot stay still for long	○	○	○
Often complains of headaches, stomach-aches or sickness	○	○	○
Shares readily with other children, for example toys, treats, pencils	○	○	○
Often loses temper	○	○	○
Rather solitary, prefers to play alone	○	○	○
Generally well behaved, usually does what adults request	○	○	○
Many worries or often seems worried	○	○	○
Helpful if someone is hurt, upset or feeling ill	○	○	○
Constantly fidgeting or squirming	○	○	○
Has at least one good friend	○	○	○
Often fights with other children or bullies them	○	○	○
Often unhappy, depressed or tearful	○	○	○
Generally liked by other children	○	○	○
Easily distracted, concentration wanders	○	○	○

	Not True	Somewhat True	Certainly True
Nervous or clingy in new situations, easily loses confidence	○	○	○
Kind to younger children	○	○	○
Often lies or cheats	○	○	○
Picked on or bullied by other children	○	○	○
Often volunteers to help others (parents, teachers, other children)	○	○	○
Thinks things out before acting	○	○	○
Steals from home, school or elsewhere	○	○	○
Gets along better with adults than with other children	○	○	○
Many fears, easily scared	○	○	○
Good attention span, sees work through to the end	○	○	○

50. **For each item, please click the box for Not True, Somewhat True or Certainly True.**

It would help us if you answered all items as best you can even if you are not absolutely certain.

Please give your answers on the basis of the child's behaviour over the last six months or this school year.

(C) Robert Goodman, 2005.

	Not True	Somewhat True	Certainly True
Considerate of other people's feelings	○	○	○
Restless, overactive, cannot stay still for long	○	○	○
Often complains of headaches, stomach-aches or sickness	○	○	○
Shares readily with other youth, for example CD's, food, games	○	○	○
Often loses temper	○	○	○

	Not True	Somewhat True	Certainly True
Would rather be alone than with other young people	○	○	○
Generally well behaved, usually does what adults request	○	○	○
Many worries or often seems worried	○	○	○
Helpful if someone is hurt, upset or feeling ill	○	○	○
Constantly fidgeting or squirming	○	○	○
Has at least one good friend	○	○	○
Often fights with other young people or bullies them	○	○	○
Often unhappy, depressed or tearful	○	○	○
Generally liked by other young people	○	○	○
Easily distracted, concentration wanders	○	○	○
Nervous or clingy in new situations, easily loses confidence	○	○	○
Kind to younger children	○	○	○
Often lies or cheats	○	○	○
Picked on or bullied by other young people	○	○	○
Often volunteers to help others (parents, teachers, other children)	○	○	○
Thinks things out before acting	○	○	○
Steals from home, school or elsewhere	○	○	○
Gets along better with adults than with other young people	○	○	○
Many fears, easily scared	○	○	○
Good attention span, sees work through to the end	○	○	○

FASD screening tool pilot study

Childhood Executive Function Inventory

Please remember to keep {{ Q16 }} in mind while answering the following questions.

51. Below, you will find a number of statements. Please read each statement carefully and thereafter indicate how well that statement is true for the child. You indicate your response by clicking one of the boxes after each statement.

My child:

	Definitely not true	Not true	Partially true	True	Definitely true
Has difficulty remembering lengthy instructions	○	○	○	○	○
Seldom seems to be able to motivate him-/herself to do something that he/she doesn't want to do	○	○	○	○	○
Has difficulty remembering what he/she is doing, in the middle of an activity	○	○	○	○	○
Has difficulty following through on less appealing tasks unless he/she is promised some type of reward for doing so	○	○	○	○	○
Has a tendency to do things without first thinking about what could happen	○	○	○	○	○
When asked to do several things, he/she only remembers the first or last	○	○	○	○	○
Has difficulty coming up with a different way of solving a problem when he/she gets stuck	○	○	○	○	○
When something needs to be done, he/she is often distracted by something more appealing	○	○	○	○	○

	Definitely not true	Not true	Partially true	True	Definitely true
Easily forgets what he/she is asked to fetch	○	○	○	○	○
Gets overly excited when something special is going to happen (e.g., going on a field trip, going to a party)	○	○	○	○	○
Has clear difficulties doing things he/she finds boring	○	○	○	○	○
Has difficulty planning for an activity (e.g., remembering to bring everything necessary for a field trip or things needed for school)	○	○	○	○	○
Has difficulty holding back his/her activity despite being told to do so	○	○	○	○	○
Has difficulty carrying out activities that require several steps (e.g., for younger children, getting completely dressed without reminders; for older children, doing all homework independently)	○	○	○	○	○
In order to be able to concentrate, he/she must find the task appealing	○	○	○	○	○
Has difficulty refraining from smiling or laughing in situations where it is inappropriate	○	○	○	○	○
Has difficulty telling a story about something that has happened so that others may easily understand	○	○	○	○	○

	Definitely not true	Not true	Partially true	True	Definitely true
Has difficulty stopping an activity immediately upon being told to do so. For example, he/she needs to jump a couple of extra times or play on the computer a little bit longer after being asked to stop	○	○	○	○	○
Has difficulty understanding verbal instructions unless he/she is also shown how to do something	○	○	○	○	○
Has difficulty with tasks or activities that involve several steps	○	○	○	○	○
Has difficulty thinking ahead or learning from experience	○	○	○	○	○
Acts in a wilder way compared to other children in a group (e.g., at a birthday party or during a group activity)	○	○	○	○	○
Has difficulty doing things that require mental effort, such as counting backwards	○	○	○	○	○
Has difficulty keeping things in mind while he/she is doing something else	○	○	○	○	○

FASD screening tool pilot study

Brief Assessment Checklist for Children

52. Here are some statements that describe children's behaviour and feelings.

For each statement, please click on the check box that best describes your child in the last 4 to 6 months.

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	Not true	Partly true	Mostly true
Can't concentrate, short attention span	○	○	○
Craves affection	○	○	○
Eats too much	○	○	○
Fears you will reject her/him	○	○	○
Hides feelings	○	○	○
Is convinced that friends will reject her/him	○	○	○
Lacks guilt or empathy	○	○	○
Prefers to be with adults, rather than children	○	○	○
Relates to strangers 'as if they were family'	○	○	○
Seems insecure	○	○	○
Startles easily ('jumpy')	○	○	○
Suspicious	○	○	○
Too dramatic (false emotions)	○	○	○
Too friendly with strangers	○	○	○
Too jealous	○	○	○
Treats you as though you were the child and she/he was the parent	○	○	○
Uncaring (shows little concern for others)	○	○	○

53. For each of the following statements please click on the check box that best describes your child in the last 4 to 6 months.

	Did not occur in the past 4-6 months	Occurred once in the past 4-6 months	Occurred more than once in the past 4-6 months
Distressed or troubled by traumatic memories	○	○	○
Does not show pain if physically hurt	○	○	○
Sexual behaviour not appropriate for her/his age	○	○	○

FASD screening tool pilot study

Neurobehavioural Screening Tool

Please remember to keep {{ Q16 }} in mind while answering the following questions.

54. The following questions aim to gain a picture of the child's behaviour within the last 6 months.

Has your child been seen, or accused of, or thought to:

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	Yes	No
Have acted too young for his or her age?	○	○
Be disobedient at home?	○	○
Lie or cheat?	○	○
Lack guilt after misbehaving?	○	○
Have difficulty concentrating, and can't pay attention for long?	○	○
Act impulsively and without thinking?	○	○
Have difficulty sitting still is restless or hyperactive?	○	○
Display acts of cruelty, bullying or meanness to others?	○	○
Thought to steal items from home?	○	○
Thought to steal items outside of the home?	○	○

FASD screening tool pilot study

Child Health Utility Index

Please remember to keep {{ Q16 }} in mind while answering the following questions

These questions ask about how your child is feeling **today.** For each question, read all of the choices and decide which one is the most like your child today. Then click on the box which best describes your child today.

Only tick one box for each question.

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55. Worried

- ◯ My child doesn't feel worried today
- ◯ My child feels a little bit worried today
- ◯ My child feels a bit worried today
- ◯ My child feels quite worried today
- ◯ My child feels very worried today

56. Sad

- ◯ My child doesn't feel sad today
- ◯ My child feels a little bit sad today
- ◯ My child feels a bit sad today
- ◯ My child feels quite sad today
- ◯ My child feels very sad today

57. Pain

- ◯ My child doesn't have any pain today
- ◯ My child has a little bit of pain today
- ◯ My child has a bit of pain today
- ◯ My child has quite a lot of pain today
- ◯ My child has a lot of pain today

58. Tired

- My child doesn't feel tired today
- My child feels a little bit tired today
- My child feels a bit tired today
- My child feels quite tired today
- My child feels very tired today

59. Annoyed

- My child doesn't feel annoyed today
- My child feels a little bit annoyed today
- My child feels a bit annoyed today
- My child feels quite annoyed today
- My child feels very annoyed today

60. School Work/ Homework (such as reading, writing, doing lessons)

- My child has no problems with their schoolwork/homework today
- My child has a few problems with their schoolwork/homework today
- My child has some problems with their schoolwork/homework today
- My child has many problems with their schoolwork/homework today
- My child can't do their schoolwork/homework today

61. Sleep

- Last night my child had no problems sleeping
- Last night my child had a few problems sleeping
- Last night my child had some problems sleeping
- Last night my child had many problems sleeping
- Last night my child couldn't sleep at all

62. Daily routine (things like eating, having a bath/shower, getting dressed)

- My child has no problems with their daily routine today
- My child has a few problems with their daily routine today
- My child has some problems with their daily routine today
- My child has many problems with their daily routine today
- My child can't do their daily routine today

63. Able to join in activities (Things like playing out with your friends, doing sports, joining in things)

- ◯ My child can join in with any activities today
- ◯ My child can join in with most activities today
- ◯ My child can join in with some activities today
- ◯ My child can join in with few activities today
- ◯ My child can join in with no activities today

FASD screening tool pilot study

Follow up survey opt-in

Thank for your participating in this study.

We are seeking participants to take part in a follow up session in 2-4 weeks.

If you would like to participate, you will receive an email with a link to a survey which will ask some of the same questions that you answered in this survey.

Your participation in the follow-up study is completely voluntary, and it is anticipated that it will take between 10 and 20 minutes to complete the follow up.

Participants in the follow up session will have the opportunity to enter a raffle to win of one of 4 x $50 gift vouchers.

* 64. Would you like to participate in the follow-up session?

- ◯ Yes, I would like to participate in the follow up
- ◯ No, I do not wish to participate in the follow up

FASD screening tool pilot study

Follow up survey opt-in

Thank you for your interest in participating in the follow-up session. Please provide an email address in the box provided below. In between 2 and 4 weeks you will be sent an email link to a second survey. The follow up survey will ask some of the same questions that you have answered today, and it is anticipated that completing the follow up session will take between 10 and 20 minutes.

65. Please provide an email address that we can send the follow up survey to

FASD screening tool pilot study

End of survey

Thank you for participating as a research participant in the present study concerning whether a newly developed tool can be used to identify children at-risk of being diagnosed with FASD. Your participation is greatly appreciated.

To enter the raffle, please follow the link below. A new survey window will open and you will be asked to provide an email address. This is to ensure that the email address you provide can be kept separate from the answers that you have provided.

Raffle entry form

If you have any questions regarding this study, please contact Stewart McDougall (Ph: +61 8 8302 2983, email: stewart.mcdougall@unisa.edu.au).

If you wish to receive a summary of the findings, please follow theinstructions

If the event that you feel distressed by your participation in this study, we encourage you to contact the services listed below.

Should you have any concerns about your child's development, we encourage you to raise your concerns with your GP

Please keep a copy of this information for your future reference by copying the page. Once again, thank you for your participation in this study

Lifeline 13 11 14
Parentline
ACT: (02) 6287 3833
NSW: 1300 130 052
QLD and NT: 1300 301 300
SA: 1800 364 100
TAS: 1300 808 178
VIC: 13 22 89
WA: 1800 654 432 or (08) 6279 1200 (metro)
SANE Mental Health helpline 1800 187 263
National Organisation for Fetal Alcohol Spectrum Disorder (NOFASD) 1300 306 238
Russell Family Fetal Alcohol Disorders Association (RFFADA) 0412 550 540

Project Researchers Feel free to contact the researchers via email or telephone using the contact details above if you wish to discuss these support services and options available to you. Please note that contacting the researcher will identify you as a participant in the study; however, your responses to the survey will remain anonymous and cannot be identified.

FASD screening tool pilot study

End of survey

Thank you for your time and interest in the research, however, for ethical reasons this research is only being conducted with people over the age of 18, and who are residing in Australia or New Zealand.

Appendix I: Item Discrimination Index Calculations.

Multiple formula exist for calculating the item discrimination index. These discrimination index can be calculated by either using an extern criterion (e.g., diagnostic group) as has been reported in the body of the chapter, or by splitting the groups based on total scores. below shows the item discrimination for the all items. Results across both indices were similar, with the item discrimination based on diagnostic group tending to be lower than that calculated by splitting the group on total scores percentages. Cappelleri, Lundy, and Hays (2014, pp. 650-651) proposed a three-step process for calculating the item discrimination index based on the total scores:

1. Partition responders who have the highest and lowest overall scores on the overall scale, aggregated across all items into upper and lower groups. The upper group can be composed of the top *x* of scores on the scale, and the lower group can be composed of the bottom *x* of scores on the scale.
2. Determine the proportion of respondents in the sample who endorse or respond to each item (or to a particular category or adjacent category groups of an item) in the upper and lower groups
3. Subtract the pair of proportions noted in Step 2.

For the purposes of this study, an item was considered endorsed if the respondent had scored either a 2 or a 3 on the items (and scores of 0 to 1 non-endorsed), and groups were defined based on the top 33% of scores, and the bottom 33% of scores. At each iteration of the item selection process, cut-points were established at 33% (low group) and 66% (high group) of the total possible scores, and participants were assigned to groups based on their scores. For the purposes of this analysis, individuals scoring between 33 and 66 of the possible scores were excluded.

Item number and content	Diagnostic groups	Possible scores
Adaptive Functioning		
Communication		
13. Appears to ignore people, or not listen to what is said	.783	.912
50. Talkative, but with little depth or coherency	.714	.719
51. Unclear pronunciation of sounds/words	.451	.531
52. Restarts sentences or words multiple times, as though they can't get the word out	.537	.652
53. Makes errors with sentence structures (leaves words out, mixes up words, errors in tense (present, past, future)	.585	.719
54. Takes longer than other children to process information and respond	.736	.906
55. Uses simpler sentences than peers	.546	.662
55. Uses simpler words, and has less variety in words (smaller vocabulary)	.570	.656
57. Has difficulty understanding jokes (e.g., laughs inappropriately, or laughs at jokes but doesn't necessarily understand them when asked)	.761	.844
59. Follows along with what is said/suggested, even if it is not true	.593	.756
60. Has difficulty talking about experiences or situations so that the listener understands (e.g., telling you about what happened during the day)	.618	.781
70. History of recurrent ear infections or hearing difficulties	.293	.386
Motor Skills		
64. Has difficulty with fine motor skills or difficulty using writing or drawing implements (e.g., crayons, pencils, pens)	.308	.412
65. Has problems with balance (e.g., struggles to stand on one leg)	.237	.375
66. Has difficulty acquiring new motor skills (e.g., slow progress in learning how to swim or ride a bike)	.523	.568
Sensory processing		
63. Does not dress appropriately for the weather	.473	.642
Sleep		
69. Has sleep problems (e. g., unpredictable sleep/wake patterns, difficulty falling asleep, waking very early in the morning, irregular naps, resists going to bed at night)	.713	.73
Social Skills		
42. Has trouble making friends with similar age peers	.474	.688
43. Has trouble keeping, or maintaining, friendships	.523	.719
44. Appears unaware of the impact of his or her actions on others	.760	.849
45. Does not appear to understand personal space	.785	.912
46. Is easily led by peers	.805	.912
49. Doesn't pick up on subtle social cues, such as body language, tone of voice, and facial expressions	.736	.875
Young for Age		
47. Acts or behaves like a younger child (e.g., immaturity, emotionally younger)	.641	.787
48. Engages more readily with younger peers	.594	.756
Cognitive Ability		
Academic Achievement		
14. Struggles with simple maths concepts (e.g., counting, arithmetic)	.618	.662
15. Struggles with complex maths concepts (e.g., multiplication, word problems)	.729	.743
16. Struggles with literacy (e.g., reading comprehension, spelling, writing)	.631	.672
17. Has difficulty understanding written information	.704	.801
18. Has difficulty understanding verbal information	.714	.844
19. Is assigned schoolwork at levels below their current year	.625	.723

General cognitive ability		
20. Appears to have difficulty with thinking or talking about things that can't be seen or touched (e.g., is dependent on learning material that can be seen or touched)	.658	.813
61. Has difficulty understanding and applying the concept of time	.713	.787
62. Has difficulty understanding and applying concepts such as now, later, before, or the days of the week	.593	.693
67. Appears to zone out or look lost in thought	.595	.750
68. Appears to have difficulty applying learning across different settings	.809	.875
Learning and Memory		
21. Has more trouble remembering to do school projects or chores	.830	.917
22. Often needs information repeated a number of times before they start to learn and remember it	.854	.897
23. Appears to forget things from one day to the next (e.g., forgets school work or rules that they knew the day before)	.856	.974
24. Has poor short-term memory (e.g., forgets what was just said to them, forgets what they were doing)	.809	.938
25. Often loses their belongings (e.g., toys, lunch box, school work)	.732	.878
27. Has difficulty anticipating the consequences of their actions	.808	.912
28. Repeats mistakes without seeming to realise or learn from previous attempts	.880	.969
29. Does not appear to care about negative consequences to their behaviour	.761	.844
41. Needs tasks broken down step by step	.831	.949
Self-regulation		
Attention		
39. Is easily distracted by other things around them	.831	.949
40. Needs more help from an adult to stay on task	.830	.923
Behavioural Regulation		
01. Talks much more than other children	.308	.343
02. Has difficulty playing or engaging in leisure activities quietly	.687	.791
03. Has more difficulty waiting for their turn than other children	.712	.824
04. Is often "on the go" or acts as if "driven by a motor"	.711	.804
05. Finds it difficult to stop doing something when asked to	.758	.923
06. Appears to act without thinking	.829	.974
07. Acts in a more hyperactive or excited way	.808	.912
08. Requires constant/or high levels of supervision	.760	.943
09. Displays more risk taking behaviour (e.g., climbs onto the roof, jumps from high places, walks onto the road without looking)	.688	.886
10. Behaviour is often inflexible, (e.g., gets "stuck" on an idea or object)	.688	.829
11. Runs about or climbs excessively when it is inappropriate	.665	.881
12. Interrupts or intrudes on others (e.g., butts into conversations or games)	.638	.809
26. Takes things which do not belong to them	.380	.531
34. Needs a lot of time to transition between activities	.640	.886
35. Is easily over-whelmed, or over-stimulated	.807	.974
37. Has difficulty adapting to changes of routines	.736	.912
38. Destroys belongings, either their own or others	.475	.625
58. Appears to replace fact with fantasy	.666	.813
Emotional regulation		
30. Has outbursts for little apparent reason	.711	.886
31. Has frequent meltdowns	.639	.861
32. Exhibits wide mood swings	.521	.730
33. Has emotional outbursts that end suddenly	.546	.662

36. Becomes agitated if they aren't getting their way	.710	.872

Appendix J: Preliminary Reliability and Validity Study Missing Data Approaches

PFST

Ten participants were missing at least one response on the 57-items retained in the PFST. These missing values were replaced with the mean score from the participants' remaining responses (Osborne, 2013). Sensitivity and specificity analyses were conducted with both the original data set, excluding participants with missing data, and the data set where missing data was replaced with the mean. The results presented below are that of the data set with the ten replaced values, and the original analysis is presented in the appendix.

CHEXI

One participant exited the survey prior to completing the CHEXI and this participant was excluded from any analyses involving the CHEXI. A further two participants were missing data on the CHEXI, both of whom skipped one question. Missing responses were replaced with mean values, as per the procedure adopted for the PFST.

SDQ

In the instructions for scoring, the test developers note that if at least three items are scored for each sub-scale, the score can be scaled up based on the three completed items. Four participants had missing data on the SDQ, none of whom had more than one missing score for any subscales and thus met the requirements for pro-rata scoring. Scale scores were replaced with the scaled-up score for analysis.

Appendix K: Skew and Kurtosis of PFST and Convergent Validity Measures

Table K

Skew and kurtosis for the PFST and convergent/divergent validation scales

	N	Skew	SE Skew	Kurt.	SE Kurt	Min	Max
PFST	83	0.02	0.26	-1.69	0.52	1	166
SDQ Emotional Problems	83	0.23	0.26	-1.14	0.52	0	9
SDQ Conduct Problems	83	0.63	0.26	-0.63	0.52	0	10
SDQ Hyperactivity	83	-0.11	0.26	-1.58	0.52	0	10
SDQ Peer Problems	83	0.73	0.26	-0.34	0.52	0	10
SDQ Prosocial	83	-0.40	0.26	-0.88	0.52	2	10
SDQ Total Problem	83	0.01	0.26	-1.36	0.52	0	34
CHEXI Working Memory Total	82	-0.17	0.27	-1.71	0.53	13	65
CHEXI Inhibition Total	82	-0.36	0.27	-1.34	0.53	12	55
BACC Total	81	0.41	0.27	-1.11	0.53	0	35
NST Total	81	0.03	0.27	-1.31	0.53	0	10

PFST- Preliminary FASD Screening Tool, SDQ- Strengths and Difficulties Questionnaire, CHEXI- Childhood Executive Functioning Inventory, BAC-C- Brief Assessment Checklist for Children, NST- Neurobehavioural Screening Tool, SE- Standard Error, Kurt- Kurtosis, Min- Minimum, Max- Maximum.

Appendix L: Formulae Used in the Calculation of Accuracy Measures.

	"Gold Standard"		
Results of new test	Present	Absent	Row total
Present	A True Positive	B False Positive	A + B
Absent	C False Negative	D True Negative	C + D
Column total	A + C	B + D	

Accuracy Indicator	Formula
Sensitivity	$\frac{A}{A + C}$
Specificity	$\frac{D}{D + B}$
Overall Accuracy rate	$\frac{(A + D)}{(A + B + C + D)}$
Positive Likelihood	$\frac{Sensitvity}{(1 - Specificity}$
Negative Likelihood	$\frac{(1 - Sensitivity)}{Specificity}$
PPP	$\frac{Prevalence \times Sensitvity}{(Prevalence \times sensitvity) + [(1 - Prevalence) \times (1 - Specifcity)]}$
NPP	$\frac{(1 - Prevalence) \times Specificity}{[(1 - Prevalence) \times Specificity)] + [Prevalence \times (1 - Sensitivity)]]}$

Appendix M: Scatterplots for convergent validation

Scatter plots showing the distribution of scores for each convergent/divergent scale are presented (Figures M.1. — M1.10.). As can be seen in the scatter plot for each measure, children with FASD are scoring higher than the community sample on most measures, with the exception of the pro-social scale of the SDQ on which children with FASD are scoring lower as this is reverse scored. On the SQD hyperactivity subscale, there appears to be a ceiling effect, with a larger number of children with FASD scoring the maximum of 10.



Figure M.1. PFST by SDQ Emotional Problems.

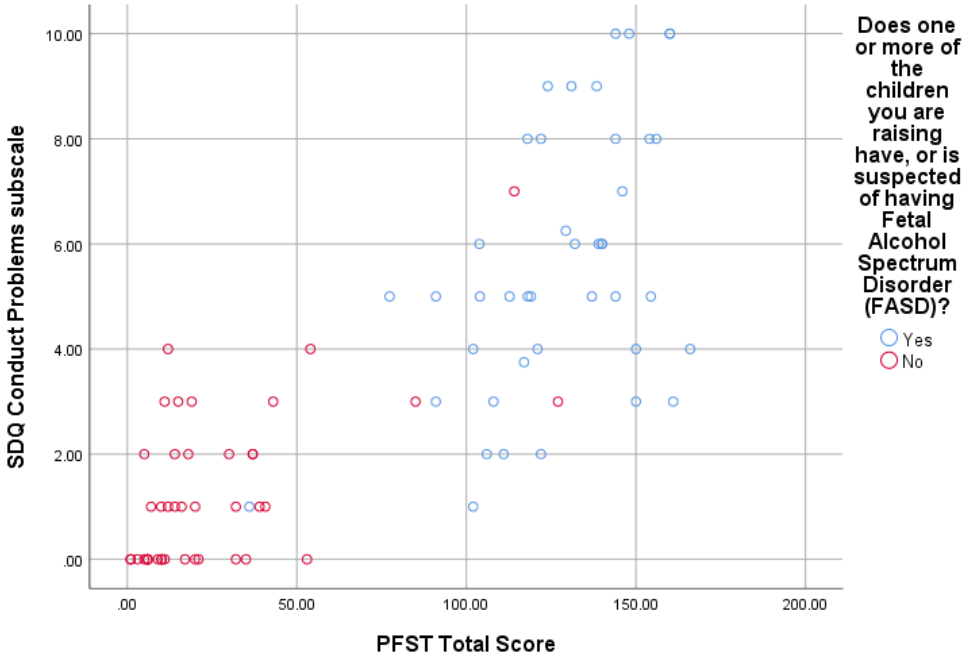


Figure M.2. PFST by SDQ Conduct Problems.

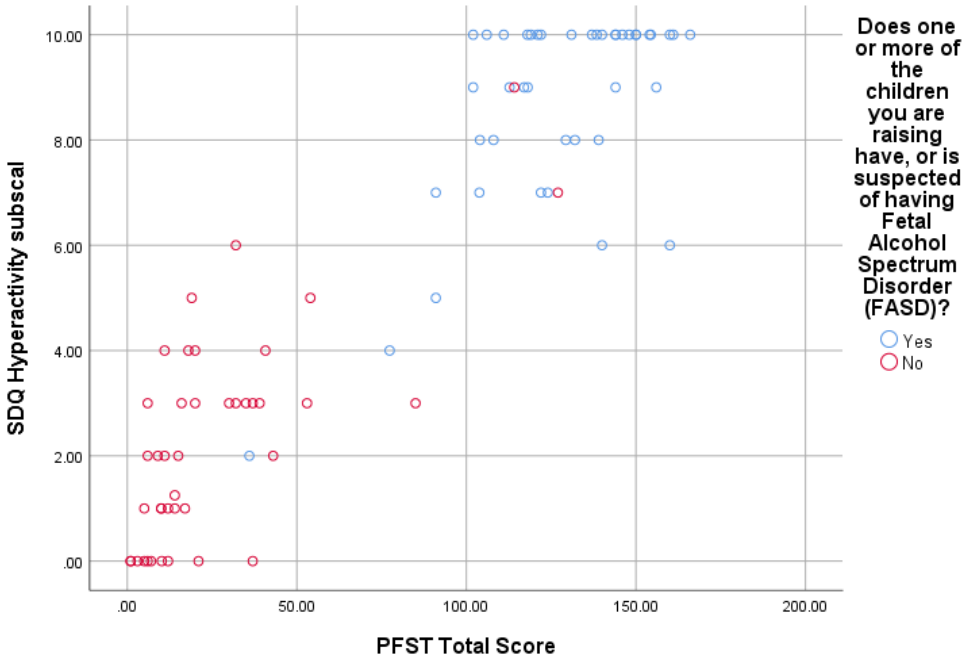


Figure M.3. PFST by SDQ Hyperactivity.



Figure M.4. PFST by SDQ Peer Problems.

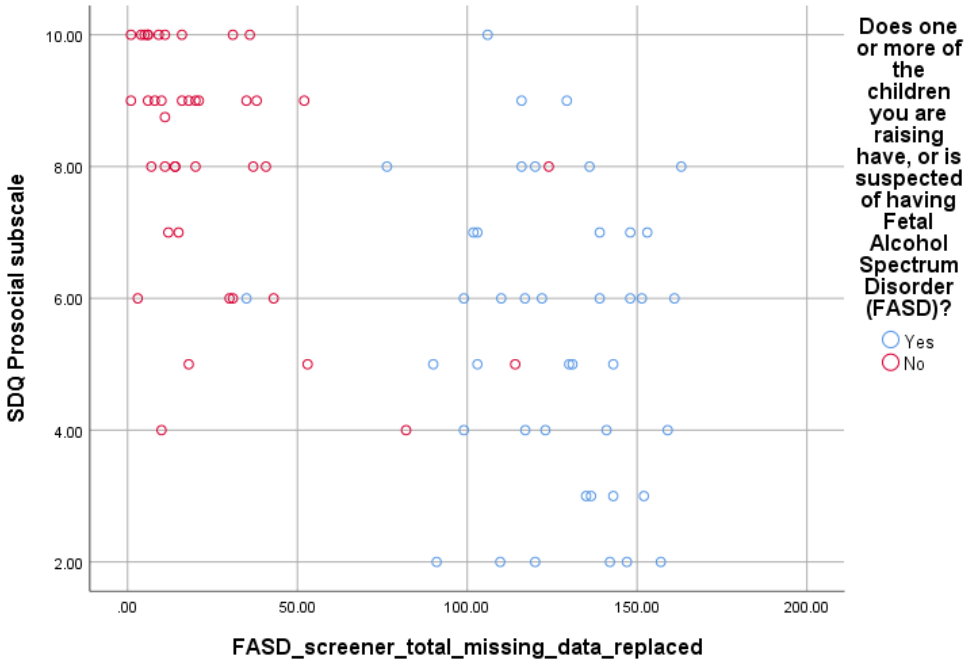


Figure M.5. PFST by SDQ Prosocial Scale.

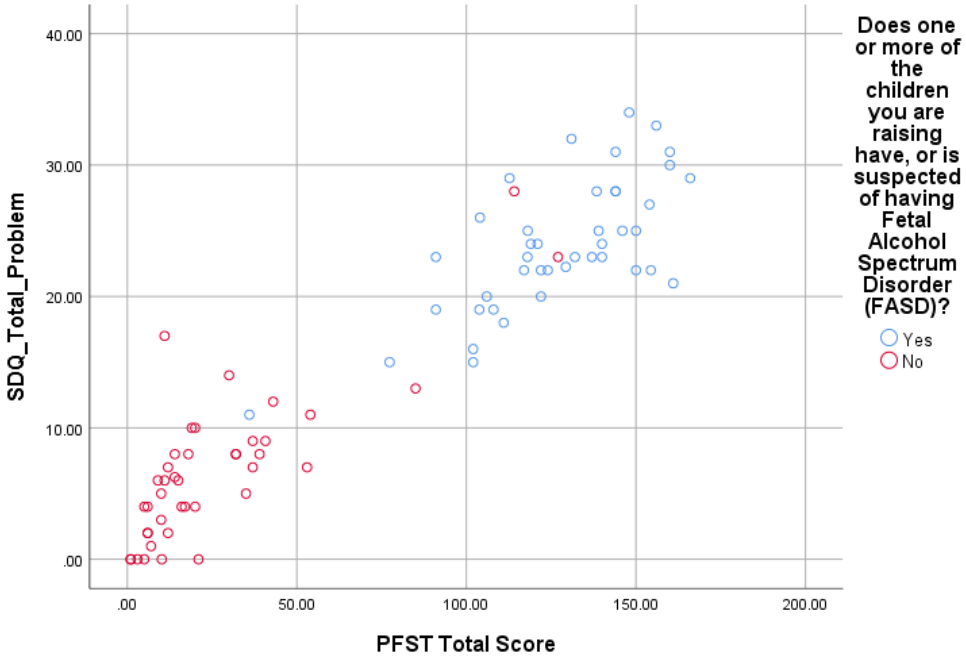


Figure M.6. PFST by SDQ Total Problem score.

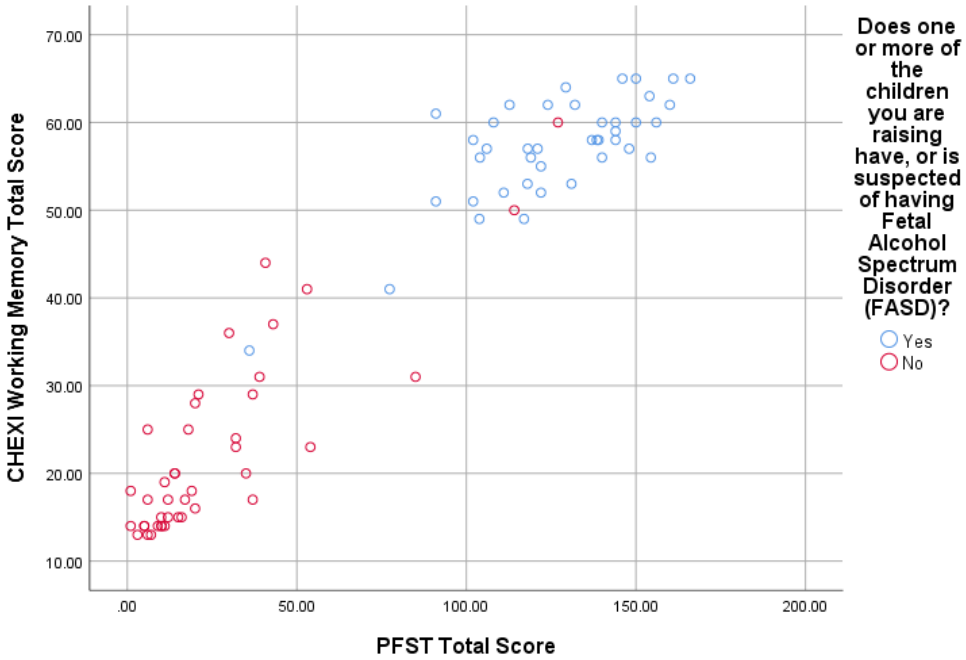


Figure M.7. PFST by CHEXI Working Memory.

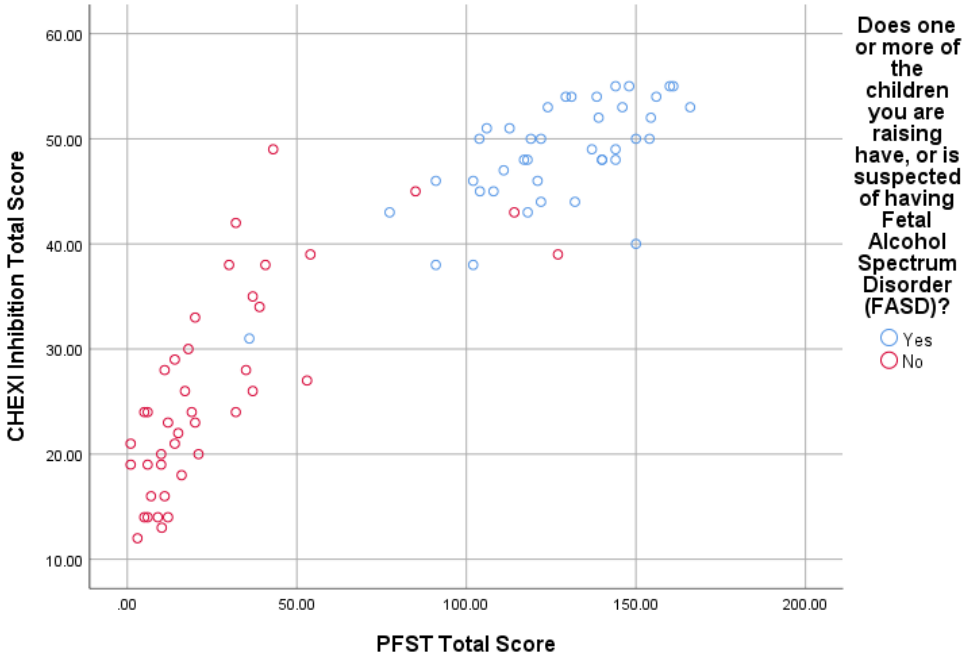


Figure M.8. PFST by CHEXI Inhibition.



Figure M.9. PFST by NST total score.

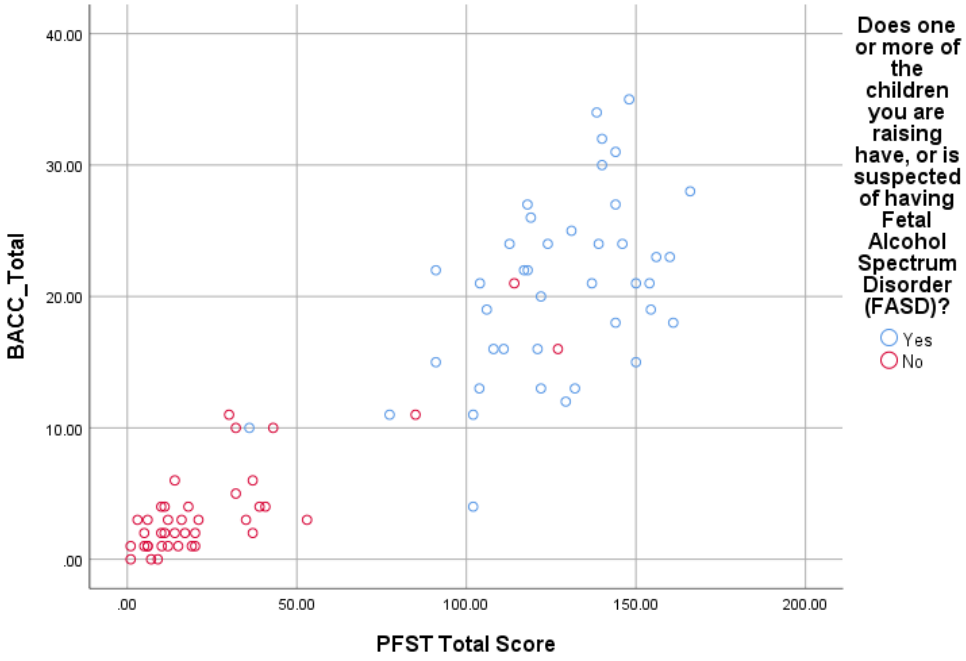


Figure M.10. PFST by BAC-C total score.

Appendix N: Sensitivity and specificity of the PFST at each possible cut-point

Table N.1.

The sensitivity and specificity of the PFST and corresponding Youden's J and minimum distance, at each possible cut-point.

PFST Score Positive if Greater Than or Equal To	Sensitivity	Specificity	Youden's J	Minimum distance
0.000	1.000	0.000	0.000	1.000
2.000	1.000	0.049	0.049	0.951
4.000	1.000	0.073	0.073	0.927
5.500	1.000	0.122	0.122	0.878
6.500	1.000	0.195	0.195	0.805
8.000	1.000	0.220	0.220	0.780
9.500	1.000	0.244	0.244	0.756
10.080	1.000	0.293	0.293	0.707
10.580	1.000	0.317	0.317	0.683
11.500	1.000	0.366	0.366	0.634
13.000	1.000	0.415	0.415	0.585
14.500	1.000	0.463	0.463	0.537
15.500	1.000	0.488	0.488	0.512
16.500	1.000	0.512	0.512	0.488
17.500	1.000	0.537	0.537	0.463
18.500	1.000	0.561	0.561	0.439
19.500	1.000	0.585	0.585	0.415
20.500	1.000	0.634	0.634	0.366
25.500	1.000	0.659	0.659	0.341
31.000	1.000	0.683	0.683	0.317
33.500	1.000	0.732	0.732	0.268
35.500	1.000	0.756	0.756	0.244
36.500	0.976	0.756	0.732	0.245
38.000	0.976	0.805	0.781	0.197

39.865	0.976	0.829	0.805	0.172
41.865	0.976	0.854	0.830	0.148
48.000	0.976	0.878	0.854	0.124
53.500	0.976	0.902	0.879	0.100
65.680	0.976	0.927	0.903	0.077
81.180	0.952	0.927	0.879	0.087
88.000	0.952	0.951	0.904	0.068
96.500	0.905	0.951	0.856	0.107
102.910	0.857	0.951	0.808	0.151
103.910	0.833	0.951	0.785	0.174
105.000	0.810	0.951	0.761	0.197
107.000	0.786	0.951	0.737	0.220
109.500	0.762	0.951	0.713	0.243
111.865	0.738	0.951	0.689	0.266
113.425	0.714	0.951	0.666	0.290
115.560	0.714	0.976	0.690	0.287
117.500	0.690	0.976	0.666	0.310
118.045	0.667	0.976	0.642	0.334
118.545	0.643	0.976	0.618	0.358
120.000	0.619	0.976	0.595	0.382
121.500	0.595	0.976	0.571	0.405
123.000	0.548	0.976	0.523	0.453
125.500	0.524	0.976	0.499	0.477
128.155	0.524	1.000	0.524	0.476
130.155	0.500	1.000	0.500	0.500
131.500	0.476	1.000	0.476	0.524
134.500	0.452	1.000	0.452	0.548
137.720	0.429	1.000	0.429	0.571
138.720	0.405	1.000	0.405	0.595
139.500	0.381	1.000	0.381	0.619
142.000	0.333	1.000	0.333	0.667
145.000	0.262	1.000	0.262	0.738
147.000	0.238	1.000	0.238	0.762

149.000	0.214	1.000	0.214	0.786
152.000	0.167	1.000	0.167	0.833
154.200	0.143	1.000	0.143	0.857
155.200	0.119	1.000	0.119	0.881
158.000	0.095	1.000	0.095	0.905
160.500	0.048	1.000	0.048	0.952
163.500	0.024	1.000	0.024	0.976
167.000	0.000	1.000	0.000	1.000

Note. PFST = Preliminary FASD Screening Tool

Appendix O: Supplementary tables accompanying scoping review

Supplementary Table O.1

Characteristics of studies comparing children with FASD to Non-exposed peers

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
Adnams et al (2001) South Africa	CON = 34, 41% F 7.12(0.5) Range = 6.3 – 8.0 FASD = 34 6.99(0.76), 41% F CON=34 4.2(1.00), 32% F Range = 3 5.5.	32.4% of FAS group living with guardians Lower maternal education in FAS group.	IoM Criteria	Active-case ascertainment methodology to identify children with FAS among children in the first grade in a school setting Children below 10th percentile in head circumference, or height and weight, referred for dysmorphology assessment	Controls recruited from same community, and included if showing no growth restriction and mothers did not report moderate or heavy alcohol consumption (not defined by authors) Control group selected to match for age, sex, first language, and home income	GMDS	Motor skills Adaptive behaviour	Covariates Maternal Education
Aragon et al., (2008) Italy	FASD = 23, 52% F 6.13(0.34) CON = 57, 35% F 6.15(0.41), Range = 6 – 7	Proportion of children in care of biological parents not reported No differences on maternal income or education	Revised IoM	Active-case ascertainment methodology to identify children with FAS or pFAS in a school setting Tiered screening; 1) less than 10th percentile on height, weight, or head size, 2) dysmorphology assessments, 3) neuropsychological testing	Controls randomly selected from same classes and received same screening as FAS/pFAS group Maternal alcohol consumption assessed in health interview, based on retrospective reports Unclear if CON group reporting alcohol use were excluded	WISC-R RCPM Rustioni Test of Language Comprehension IPDA DBD	Academic achievement Attention Executive functioning Language	Covariates: none
Breiner et al., (2013) Canada	FASD = 17 Deferred = 18 CON = 25 Range =4 – 6		Canadian guidelines	Recruited from children referred to the Motherisk clinic for FASD diagnostic assessment	Limited information provided. States that "typically developing normal control children who participated as controls in	NST	FASD Behaviour	Assessments conducted using the CBCL, and then transformed to the NST scores Covariates: none

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	Mean age and SD not reported, gender not reported				neurodevelopmental follow up studies at Motherisk"			
Chen et al., (2012) USA	FASD = 33 7.5(2.2), 33% F CON = 418, 7.2(2.7), 50% F Range= 4 – 12	70% of FASD group in foster or adoptive care ,18% living with one or both birth parents, 12% not reported CON not reported	4-Digit Diagnostic Code	Recruited from FAS DPN subject pool. Included if seen by multidisciplinary assessment team, received a formal FASD diagnosis and if enrolled in behavioural intervention study Excluded if seizure disorder requiring daily medication, IQ < 55, diagnosed thought disorder or very serious conduct problems	CON group recruited from 3 public elementary schools CON group exclusion unclear, maternal alcohol use not assessed	CSHQ Sleep logs	Sleep	Note: CON group used re-analysis of existing data, not new data collected for this study FASD group had higher proportion male, less paternal presence in household and lower SES Covariates: none
Coles et al., (1997) USA	FAS/FAE = 25 7.46(0.41), 44% F ETOH =62 7.64(0.49), 61% F ADHD = 27 7.68(0.54), 19% F CON = 35 7.73(0.54), 51% F Range = 7 – 8.5	Not reported, states in discussion that the FAS/FAE group "for the most part, had not experienced highly disorganised environments or multiple custody arrangements", but not quantified	FAS/FAE; alcohol exposed and display 1+ sentinel facial feature ETOH; alcohol exposed, no dysmorphology	Longitudinal follow up of study of prenatal exposure	CON group those who reported no alcohol use in pregnancy ADHD group recruited from diagnostic clinics	K-ABC WISC- R WCST Kisborne Paired Associates Test VMI Vigilance Task Performance CPT SNAP-IV CBCL	Academic achievement Attention Affect regulation Executive functioning Motor skills	Few children with FAS were able to complete the CPT, potentially skewing results Covariates: none
Davies et al., (2017) South Africa	FAS/pFAS = 29 62% F PAE (No FASD) = 57 54% F CON= 35, 63% F Mean age not reported	Not reported	Revised IoM	Follow up of children born in region hospital between 2002 and 2003. FAS assessment conducted at age 5, with prenatal alcohol collected in pregnancy	Assigned to control group if no reports of alcohol exposure during pregnancy	GMDS Locomotor, Personal-social, Language, Eye-hand coordination, Performance, Practical reasoning and Total score	Adaptive behaviour Motor skills Language and communication	Covariates: none

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	Range= 5 – 6							
Doney, Lucas, Jirikowic et al., (2017) Australia	FASD = 21 PAE (No FASD) = 39 CON = 39 Ages for subgroups not reported. Total sample = 8.7 Range = 7.5 – 9.6	Not reported	Lililwan Study	Population prevalence study	Controls from same cohort, and included if not meeting the criteria for FASD	ETCH Sentence composition task Miller Function and Participation Scales: Draw-a-Kid- game	Motor skills	Covariates: none
Doney et al., (2016) Australia	FASD = 21 PAE (No FASD) = 39 CON = 39 Ages for subgroups not reported. Total sample = 8.7 Range = 7.5 9.6	Not reported	Lililwan Study	Population prevalence study	Controls from same cohort, and included if not meeting the criteria for FASD	VMI Scores reported: Total VMI score, and scores for Visual Perception and Motor Coordination subscales	Motor skills	Mean VMI scores across the whole sample were in the below average range, which may reflect the high level of adversity and presence of multiple neurodevelopmental risk factors Covariates: none
Doney, Lucas, Watkins et al., (2017) Australia	FASD = 21 PAE (No FASD) = 39 CON = 39 Ages for subgroups not reported. Total sample = 8.7 Range =7.5 9.6	Not reproted	Lililwan Study	Population prevalence study (Lililwan study)	Controls from same cohort, and included if not meeting the criteria for FASD	BOT-2 .	Motor skills	Covariates: none
Fuglestad et al., (2015) USA	FASD = 39 4.4(0.76), 67% F CON=50 4.2(1.00), 32% F Range = 3 5.5.	97% of the FASD group in adoptive or foster care.	Revised IoM	FASD group Recruited from attendees to university clinics	Control group recruited from participant database (families who gave birth to a child at any area hospitals were invited to be contacted for future university research)	EF Scale for Children Delay of Gratification Task	Executive functioning	Covariates: Age
Greenbaum et al., (2009) Canada	FASD = 33 9.23, 52% F ADHD = 30 9.33, 20% F	82% of the FASD group in adoptive or foster care, 15% in care with access to biological mother, 3%	Diagnostic criteria not reported Children complete battery of tests	FASD group recruited from diagnostic clinic records.	ADHD group recruited form postings in pediatric clinics, or pre-existing data pool	CBCL SSRS Test of Social Cognition MNTAP	Adaptive behaviour, Social skills and social cognition	Covariates: SES, Abuse history, Performance IQ

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	CON = 34 8.87, 47% F Range = 6 13 Age SD not reported	in care of biological relative.	and decision made based on % of "deficits and assets"		CON group recruited from community postings, district school board, or the non-exposed foster or adoptive siblings of children with FASD ADHD and CON group excluded if history of exposure to alcohol or other teratogens, or documents learning disability or medical disorder affecting brain function.		Affect regulation	
Hansen & Jirikowic (2013) USA	FASD = 11 8.75(2.16), 45% F CON = 12 8.25(2.25), 42% F Range = 5 11	45% of the FASD group in adoptive care or in care of legal guardians. 55% of FASD group in care of birth parents. All CON group in care of birth parents.	4-Digit Diagnostic Code	FASD group recruited from clinical database (FAS DPN)	Control group recruited from university research participant pool, and word of mouth	SSP SPM	Sensory processing	Covariates: none
Jirikowic, Kartin et al., (2008) USA	FASD = 25 6.5(0.88), 44% F CON = 23 7.0(0.95), 52% F Range= 5 8.	92% of FASD group in foster or adoptive care. 8% in care of biological parents. 79% of CON group in care of biological parents, 9% in foster care. Not reported = 12%	4-Digit Diagnostic Code	FASD group recruited from clinical database	Control group recruited through mailing to parents children in local kindergartens, schools, and after-school programs.	SIBR	Adaptive behaviour Social skills and social communication.	Covariates IQ scores from TONI
Jirikowic, Olson et al., (2008) USA	FASD = 25 6.5(0.88), 44% F CON = 26 6.9(0.85), 46% Range = 5 8.	92% of FASD group in foster or adoptive care. 8% in care of biological parents. 74% of CON group in care of biological parents, 4% in foster care. Not reported = 23%	4-Digit Diagnostic Code	FASD group recruited from clinical database	Control group recruited through mailing to parents children in local kindergartens, schools, and after-school programs.	SSP BOTMP QNST-II NEPSY Sensimotor Core NEPSY Visuospatial Processing Core TONI-2 SIB-R WRAT-3 SSRS	Academic achievement Adaptive behaviour, Social skills, and social communication Motor skills Sensory processing	Covariates IQ scores from TONI

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
Kalberg et al., (2013) South Africa	FASD = 61 6.54(0.63), 54% F CON = 52 7.36(0.92), 56% Range not reported.	Report that 53 of 61 biological mothers were available for interviews	Revised IoM	Population prevalence study	Control group recruited some pool of consenting first grade students who was not reported to have been exposed to alcohol prenatally.	PCBL-36 RCPM TROG VMI JSAIS Digit Span, Absurd Situation, Word Association, and story memory subtests	Executive function Language Memory Motor skills FASD-specific behaviour	Covariates Age
Kodituwakku, Adnams et al., (2006) South Africa	FAS = 62 7.63(0.91), 53% F CON = 61 7.55(0.87), 52% F Range not reported.	Not reported	Revised IoM	Recruited through two-step active case ascertainment study; screening for height, weight, and head circumference >10 percentile and follow up dysmorphology assessment.	CON selected based on minimal exposure as retrospectively reported by parents.	RCPM TROG Modified Children's Test of Non-word Repetition Letter Fluency Category Fluency	Executive function Language	Covariates Raven's Progressive Matrix scores
Kodituwakku, Coriale et al., (2006) Italy	FASD = 22 6.3(0.46), 50% F CON = 60 6.2(0.38), 52% F Range = 6.1 7.7	Not reported	Revised IoM	Enrolled in first grade at randomly selected schools, active case recruitment	Matched controls selected from children in participating school, included if reported to have not been prenatally exposed to alcohol.	RCPM Rustioni Test of Language Comprehension IDPA DBD PBCL-36	Academic achievement Attention Executive function Language FASD-specific behaviour	Estimated number of drinks during pregnancy did not differ between groups. Covariates None
Kooistra et al., (2010) Canada	FASD = 30 8.83(1.17), 43% F ADHD = 47 9.25 (1.67), 34% F CON = 39 9.1(1.08), 49% F Range = 7 10.	Not reported	4-Digit Diagnostic Code	All children aged 7 10 attending FASD clinic at pediatric hospital invited to participate	Control group recruited from two elementary schools in local region via posters and parents' councils.	CPT Go/No-Go Task WISC-III	Attention Executive Function	Covariates: none
Kooistra et al., (2011)	FASD = 28 8.81(1.25), 43% F	Not reported	4-Digit Diagnostic Code	All children aged 7-10 attending FASD clinic at	Control group recruited from two elementary schools in local region	ANT WISC-III	Attention	Covariates Age, gender, SES, and estimated FSIQ

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
Canada	ADHD = 47 9.08(1.93), 32% F CON = 38 9.13(1.08), 47% F Range= 7 10			pediatric hospital invited to participate	via posters and parents' councils.			
Kooistra et al., 2009 Canada	FASD = 30 8.8(1.2), 43% F ADHD = 47 9.3(1.7), 34% F CON = 39 9.1(1.1), 49% F Range = 7 10	Not reported	4-Digit Diagnostic Code	All children aged 7-10 attending FASD clinic at pediatric hospital invited to participate	Control group recruited from two elementary schools in local region via posters and parents' councils.	M-ABC COMPS	Motor Skills	Covariates: none
Kully-Martens et al., (2012) Canada (Pooled)	FASD = 19 9.05, 58% F CON = 38 8.97, 53% F SD not reported Range = 6 12	74% of FASD group in adoptive or foster care. 26% of FASD group in care of biological parents. 100% of CON group in care of biological parents.	Either Canadian Diagnostic Guidelines or 4-Digit Diagnostic Code	FASD group recruited from FASD clinical services division of hospital	CON group through local elementary school, screened for neurological condition and PAE	Recognition memory performance Source memory performance	Memory	Covariates: none
Lane et al., (2014) Canada (Pooled)	FASD = 14 11.73 (1.36), 64% F CON = 14 8.42 (1.39), 64% F Range = 8 13	72% of FASD group in adoptive or foster care. 28% in care of biological father or other relative CON group not reported.	Canadian guidelines	Recruited from FASD diagnostic clinic in BC .	Control group recruited through community postings, school contacts, and distribution of flyers to acquaintances and colleagues	TEACH	Attention	Control group matched to mental age of children with FASD, based on performance on the Leiter-R non-verbal intelligence scale. Covariates: none
Lewis et al., (2016) South Africa	FAS/pFAS = 29 11.0(0.05), 45% F PAE, no FASD = 32 11.2 (0.4), 40% F CON = 28	Not reported	Revised IoM	Children participating in a follow up of Cape Town Longitudinal Study	Controls recruited from same cohort, if reported that PAE was "no more than minimal"	Prospective Memory Task EF tasks (measured, but used as a moderator)	Memory	Covariates On-going task absorption (attention) used as a co-variate in analysis of prospective memory

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	10.9 (0.3), 50% F Range = Not reported							
Lindinger et al., (2016) South Africa	FAS = 8 10.6(0.4), 63% F pFAS = 19 11.1(0.4), 42% F PAE = 17 11.1(0.4), 41% F CON = 19 11.0(0.3), 52% F Range = 9 11.	Not reported	Revised IoM PAE group-heavily exposed but not meeting FAS or pFAS criteria	Cohort of pregnant women recruited, follow up at age 3-6 for FASD diagnosis	Women who reported abstaining or drank minimally were recruited as controls.	University of Cape Town Theory of Mind NEPSY-II contextual, and affect regulation subtests RMET-C	Social skills and social communication	Minimal drinking defined as <0.5oz AA/Day per day and no binge episodes. Covariates: none
Lucas, Doney, Latimer et al.,(2016) Australia	FASD = 21 CON = 87 Ages not provided for subgroups. Mean age = 8.7 (0.6) Range = 7.5 9.6	Not reprted	Lililwan Study	Population prevalence study	Controls from same cohort, and included if not meeting the criteria for FASD	BOT-2 Gross Motor outcomes	Motor skills	Covariates: none
Lucas, Latimer, Fitzpatrick et al., (2016) Australia	FASD = 21 CON = 87 Ages not provided for subgroups. Mean age = 8.7 (0.6) Range = 7.5 9.6	Not reprted	Lililwan Study	Population prevalence study	Controls from same cohort, and included if not meeting the criteria for FASD	QNST-II	Motor skills	Covariates: none
May, Baete, Russo et al., (2014) USA (Pooled)	FAS = 12 6.96(0.54), 50% F pFAS = 23 6.98(0.44), 52% F ARND = 13 7.01(0.27), 46% F CON = 168 6.91(0.44), 44% F	Not reported	Revised IoM	Active case ascertainment based on tiered screening process. Oversampled < 25th percentile of height, weight, or OFC teacher referral available,	Controls randomly selected from same population as FASD group. Included as control if no FASD diagnosis and complete data.	VMI DAS VABS	Adaptive behaviour Social skills, and social communication Executive function Motor skills	Covariates: none Age range not reported, but all first-grade students.
May, Blankenship, Marais et al., (2013)	FAS = 68 7.12(0.73), 50% F pFAS = 52 6.78(0.79), 52% F	Not reported	Revised IoM	Active Case ascertainment Tiered screening process	Controls identified from same population, and randomly selected into either PAE or CON	TROG RCPM Digit Span TRF	Attention Executive function	Covariates: none FASD groups analysed separately

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
South Africa	ARND = 35 7.00(0.66), 49% F PAE = 38 6,73(0.56), 50% F CON = 90 6.67(0.52), 46% F			1) < 25th height, weight, head circumference percentile, 2) Dysmorphology exam, 3) Cognitive and behavioural testing	based on maternal report of PAE.		Language	Age range not reported, but all first-grade students.
May, de Vries, Marais et al., (2016) South Africa	FAS = 173 7.22(0.8), 54% F pFAS = 109 6.94(0.71), 45% F ARND = 59 7.11(0.73), 51% F CON = 212 6.68(0.64), 53% F	Not reported	Revised IoM	Active case ascertainment Tiered screening process 1) < 25th height, weight, head circumference percentile, 2) Dysmorphology exam, 3) Cognitive and behavioural testing	Controls selected from same population.	TROG RCPM Digit Span TRF total score and inattention score	Attention Executive function Language	Covariates: none FASD groups analysed separately Age range not reported, but all first-grade students.
May, de Vries, Marais et al., (2017) South Africa	FAS = 129 7.21(0.84), 47% F pFAS = 100, 56% F 7.09(0.73), ARND = 55 7.12(0.75), 47% F CON = 104 6.97(0.89), 46% F	Not reported	Revised IoM	Active case ascertainment Tiered screening process 1) < 25th height, weight, head circumference percentile, 2) Dysmorphology exam, 3) Cognitive and behavioural testing	Controls selected from same population.	TROG RCPM WISC-IV Digit Span Achenbach TRF	Attention Executive function Language	Covariates: none FASD groups analysed separately
May, Gossage, Marais et al., (2007) South Africa	FAS = 55 7.81(0.86), 42% F pFAS = 18 7.63(0.55), 39% F CON = 145 7.29(0.52), 52% F	20% of FASD group reported to be in foster or adoptive care, 33.3% of pFAS group in foster or adoptive care. 5.3% of CON group in foster or adoptive care.	Revised IoM	Active case ascertainment First-grade students from 21 schools all screened for height, weight and head circumference <10th percentile	Controls selected from same population	TROG RCPM PBCL-36	Language FASD-specific behaviour	Covariates: none FASD groups analysed separately
May, Keaster, Bozeman et al., (2015)	FAS = 7 7.13(0.48), 71% F	Not reported	Revised IoM	Active case ascertainment	Assigned to CON if not FAS, pFAS or other known anomaly.	WASI WISC-IV Digit Span, Coding, Mazes	Academic achievement Adaptive behaviour	Covariates: none

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
USA FASD analysed separately	pFAS=19 7.45(0.53), 37% F CON=186 7.04(0.37), 36% F			First-grade students all screened for height, weight and head circumference <10th percentile, or referred by teacher over behavioural concerns.	39% of CON group reported alcohol consumption during index pregnancy but noted only 1 mother reported drinking in 2nd and 3rd trimester.	WRAT-R VABS PBCL-36	FASD-specific behaviour Executive functioning	FASD groups analysed separately
May, Marais, de Vries et al., (2016) South Africa	FAS = 69 6.93(0.63), 52% F pFAS = 91 6.65(0.53), 46% F ARND = 39 6.69(0.64), 38% F CON = 207 6.52(0.53), 47% F	Not reported	Revised IoM	Active case ascertainment Tiered screening process 1) < 25th height, weight, head circumference percentile, 2) Dysmorphology exam, 3) Cognitive and behavioural testing	Assigned to CON if not FASD and completed data available 41% of CON group reported alcohol consumption during index pregnancy. Lower rates of binge drinking compared to case groups	TROG RCPM WISC-IV Digit Span Scaled score TRF Total Problems and Attention	Attention Cognition Executive functioning Language	Covariates: none FASD groups analysed separately
Olswang et al., (2010) USA FASD Pooled	FASD *n* = 12, 50% F Age = 9.2(1.1) CON *n* = 12, 50% F 9.1(1.3) Range=7-12	92% of FASD group reported to be in adoptive or foster care, 8% in care of biological parents. 8% of CON group in care of adoptive parents, 82% in care of birth parents. 8% missing	4-Digit Diagnostic Code	FASD group identified from clinical database and included only if clinical range for SSRS problem behaviours, IQ > 85 on K-BIT and in mainstream class Excluded in no classroom match available	CON group selected by teacher, matched by age (+/-6 months), gender, classroom, not in clinical range of SRSS, IQ no more than 1SD below mean (>85)	Social Communication Coding System	Social skills and social communication	Time spent engaged in classroom behaviour; Observer rating of time spent engaged in 6 behaviours as per the SCCS Changeability; how often a child moves from one of the 6 SCCS behavioural dimensions to another Stability; time spent in the 3 most frequent behaviours (Prosocial/engaged, passive/disengaged and irrelevant) across days Covariates: none

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
Kjellmer and Olswang, (2013) USA	(Re-analysis of the Olswang et al data above)	As above	4-Digit Diagnostic Code	As above	As Above	As above	As Above	Re-analysis of data reported in Olswang et al., 2010. Covariates: none
Pei et al., (2011) Canada	FASD *n* = 35, 51% F Age = 8.29 (NR) CON *n* = 35, 51% F 8.77 (NR) Range= 6-12 SD not provided.	74.2% FASD group in adoptive, foster, or kinship care. 25.8% biological, or biological and kinship 100% of CON group in care of biological parents.	Canadian guidelines	FASD group recruited from FASD Clinical Services division, Glenrose Rehabilitation Hospital	CON group recruited from local elementary school and selected based on closely of match to age and gender to FASD cases. Excluded if reported to have neurological disease or brain injury and if alcohol consumption of smoking during pregnancy reported.	Rey-Osterrieth Complex Figure	Executive functioning	Covariates: none 4 -digit diagnostic code Postnatal risk score of 3 assigned to 8 (22.9%) cases and 4 assigned to 21 (60% of cases). No risk assigned to 6 (17.6%) of cases.
Quattlebaum & O'Connor (2013) USA FASD Pooled	FAS *n* = 10, 20% F Age = 8.18(2.10), pFAS *n* = 43, 44% F Age = 8.68(1.49) ARND *n* = 44, 57% F Age = 8.60(1,47) CON *n =28*, 46% F Age = 8.30(1.43)	Not reported.	4-Digit Diagnostic Code	Recruitment through health care providers, YMCAS, and schools. PAE determined in screening interview Included if IQ >70, and 1SD below the mean of the VABS socialization subscale.	CON group formed from those reporting no PAE. Unknown exposure excluded.	BRIEF Children's Colour Trials WISC-III Digit Span and Spatial Span Test of Language Competence Figurative Language subtest VABS PBCL-36 SNAP-IV CBCL TRF Pictorial Depression Scale Hostile Attribution Test	Adaptive behavior, social skills, and social communication Affect regulation Attention Executive functioning Language	FASD groups pooled due to lack of differences between diagnostic subgroups. Demographic characteristics reported separately. Children with unknown PAE excluded. Covariates: IQ
Rasmussen & Bisanz, (2011) Canada FASD Pooled	FASD *n* = 21, 52% F Age = 5.67(NR), CON *n* = 20, 65% F Age = 5.08(NR) Range=4-6 SD not provided	90.5% of FASD group in adoptive, foster or kinship care. 9.5% in care of biological aprent. CON all biological	4-Digit Diagnostic Code	FASD group recruited through FASD diagnostic clinics, FASD education programs, First Nations communities, and FASD community agencies.	Comparison group local preschools, kindergartens, and day cares Eligibile if not diagnosed with FASD, learning disability, mathematics disability,	WMTBC WJ-III	Academic Achievement Executive Function	Covariates: none .

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
					neurodevelopmental disorder or brain injury.			
Rasmussen, Wyper et al., (2009) Canada	FASD *n* = 25, 44% F Age = 6.39(1.19), CON *n* = 28, 50% F Age = 6.32(0.88) Range=4-8	92% of FASD group in care of adoptive, foster, or kin cares. 8% in care of biological parents.	4-Digit Diagnostic Code	FASD group recruited through FASD clinics and community agencies.	CON group recruited in another city, from database of research participants. Inclusion criteria not specified.	First-order false belief task Whispers task Day/Night task WMTB-C digit recall, and block recall PPVT-III	Adaptive behavior Executive function Language	Covariates: none
Rasmussen, Talwar, Loomes et al., 2008 Canada	Preschool FASD *n* = 13, 46% F Age = 5.63(0.80), CON *n* = 12, 50% F Age = 5.51(0.78) Early elementary FASD *n* = 10, 41% F Age = 6.95(0.57), CON *n* = 12, 50% F Age = 7.48(0.83) Range= 4 to 8	Not reported	4-Digit Diagnostic Code	FASD recruitment through diagnostic clinicians and community agencies.	CON group through research participant database and eligible to participant if no other developmental disability.	Lie-Telling Paradigm	Social skills, and social cognition	Covariates: none Groups split in preschool and school aged**
Rasmussen, Pei et al., (2009) Canada FASD Pooled	Young FASD *n* = 20, 45% F Age = 5.8(NR), CON *n* = 20, 55% F Age = 5.58(NR) Older FASD *n* = 15, 33% F Age = 9.92 (0.57), CON *n* = 15, 53% F Age = 9.17(0.83)	63% of FASD in either foster or adoptive care. Remaining 27% reported to be in care of biological parent, or grandparent 91% of CON group in care of biological parents, 9% missing data.	4-Digit Diagnostic Code	FASD group recruited through FASD clinics and community agencies.	Recruitment strategy for CON group not stated. Eligible if not diagnosed with a FASD or any other neurodevelopmental disability.	Memory Strategy Task Day/Night Task PPVT-III.	Executive functioning Language Memory	Participants divided in two groups; younger (5-6, n=40) and older (8-10, n=30). Covariates: none
Stevens et al., (2015)	FASD *n* = 37, 54% F Age = 10.03 (1.4)	70% of FASD group reported to be foster or	either the Canadian	FASD group recruited through FASD clinics	CON group recruited through community	CBCL SSIS	Social skills and social communication.	Covariates: Age, Gender

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
Canada	CON *n* = 21, 52% F Age = 10.54(1.2) Range=8-12	adoptive care, 27% in care of relatives, 3% in care of biological parents. 10% of CON group in care of adoptive parents, 90% in care of biological parents.	Diagnostic guidelines (2005) or 4-Digit code system	and community agencies.	postings, or foster or adoptive siblings CON excluded if previous neurodevelopmental or behavioural diagnosis, IQ below 70 or gestational exposure to alcohol or other substances of abuse.	NEPSY-II Theory of Mind subtest Test of Social Cognition	Affect regulation. Executive functioning	
Stevens et al., (2016) Canada FASD Pooled	FASD *n* = 35, 46% F Age = 9.88 (1.3) CON *n* = 21, 48% F Age = 10.54(1.2) Range=8-12	71% of FASD group reported to be in foster or adoptive care, 26% in kinship care, and 3% in care of biological parents. 10% of CON group in care of adoptive parents, 90% in care of biological parents	Either the Canadian Diagnostic guidelines (2005) or 4-Digit code system	FASD group recruited through FASD clinics and community agencies.	CON group recruited through community postings, or foster or adoptive siblings CON excluded if previous neurodevelopmental or behavioural diagnosis, IQ below 70 or gestational exposure to alcohol or other substances of abuse.	RMET-C	Social skills, and social communication	Covariates Age, Gender
Stevens et al., 2013 Canada FASD Pooled	FASD *n* = 25, 48% F Age = 10.3 (1.5) CON *n* = 17, 47% F Age = 10.2(1.2) Range= 8 to 12	76% of FASD reported to be in care of foster or adoptive parents. 24% in kinship care 12% of CON group in adoptive care, 6% in kinship care, and 82% in care of biological parents	Either the Canadian Diagnostic guidelines (2005) or 4-Digit code system	FASD group recruited through FASD clinics and community agencies.	CON group recruited through community postings, or foster or adoptive siblings Excluded if psychiatric diagnosis, learning disability, neurological or brain abnormality, or prenatal exposure to any teratogen.	SSIS	Social Skills and social communication	Covariates FSIQ
Thomas et al., (1998) USA Pooled	FASD *n* = 15, 73% F Age = 10.3 (2.0) CON *n* = 15, 53% F Age = 10.2(1.2) IQ matched *n* = 15, 67% F	Among the FASD group, 47% reported to be in foster or adoptive care, 53% in care of birth parents. 87% of CON group in care of biologica parents.	Traditional	FAS identified through FAS Clinic VIQ referred by school psychologists and pecial education teachers	CON recruitment not detailed	WISC-III, or WISC-R VABS- social skills subscale	Adaptive behavior,	Covariates: SES

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	Age = 9.2(1.6) Range= 5 to 13							
Thorne (2017)	FASD *n* = 69, 49% F Age = 9.8 (NR) CON *n* = 69, 39% F Age = 9.6(NR) Range= 7 to 12	Not reported	4-Digit Diagnostic Code	Recruited from University of Washington (UoW) FASD Clinic.	CON group recruited from two school districts, and invited based on judgement of "typically developing" based on academic and behavioural records.	Analysis of narrative production task; grammatical errors.	Language	Covariates: none
Tsang et al., 2017 Australia	FASD=18 CON=79 Ages not provided for subgroups. Mean age total= 8.7(0.6) Range=7.5-9.6	Not reported	Lililwan Study	Lililwan study		CBCL	Executive Functioning Affect Regulation	Covariates: none
Viljoen et al., (2005) South Africa	FASD *n* = 64, 53% F Age = 6.54 (0.64) CON *n* = 146, 52% F Age = 6.36(0.55) Range= 7 to 12	17.5% of the FASD group were reported to be in foster or adoptive care 10.7% of CON group reported to be in foster or adoptive care	Revised IoM	Two tiered screening of population. Tier 1: Height, Weight, OFC at or below 10th percentile Tier 2: Complete physical	Controls selected from same population. Eligible as control if not screened through on height, weight or OFC or on dysmorphology.	RCPM TROG PBCL-36	Language Cognition FASD-Specific Behaviour	Covariates: none
Wengel et al., (2011) Canada	FASD *n* = 19, 58% F Age = 4.92 (NR) CON *n* = 12, 66% F Age = 4,33(NR) Range=3-6 SD not provided	Not reported	Canadian	FASD children recruited from FASD community organisations or outreach teams.	CON group recruited from day cares and pediatricians offices	CSHQ Sensory Profile	Sensory processing Sleep	CSHQ Sleep onset delay and daytime sleepiness subscales were omitted from the results section Covariates Age, Gender
Wyper & Rasmussen, (2011) Canada	FASD *n* = 27, 37% F Age = 9.0 (NR) CON *n* = 23, 39% F Age = 8.75 (NR)	48% of FASD group reported to be in foster or adoptive care. 19 % in kinship care, and 22% in care of biological parents.	4-Digit Diagnostic Code	FASD group recruited through hospital diagnostic clinic.	CON group recruitment lacking details. Stated as recruited "through colleagues".	CREVT-2 TOLD.	Language	Covariates: none

Author, (Year) Country	Child Demographics N Mean age (SD), % Female Range	Family demographics	FASD diagnostic criteria	FASD group Recruitment	Control group recruitment	Measures/Test reported	Domains assessed	Comments
	Range=5-13 SD not provided	CON All biological.						

Supplementary Table O.2

Characteristics of studies comparing children with PAE to Non-exposed peers

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Family Characteristics	PAE source	Recruitment	FASD criteria assessed or diagnoses?	Measures/Test reported	Domains assessed	Notes
Crocker et al., (2015) USA	PAE *n* = 29, 35% F Age = 10.02(1.86) CON *n* = 29, 48% F Age = 10.08 (1.77) Range=7-12	Alcohol exposed (PAE) defined as 4+ drinks per occasion at least once a week, or 14 drinks per week on average during pregnancy.	13.4% of PAE group in care of biological parents, 96.3% of CON.	PAE group primarily through multi-source collateral report Direct report by the caregivers (primarily biological parents) in the CON group.	Recruited as part of larger study of the "behavioural teratogenicity" of alcohol Both groups recruited through professional referral, community outreach and self-referral	Assessed by dysmorphologist for the facial features and physical measurements (i.e., height and weight). 41% of PAE group met criteria for FAS.	WRAT-R Arithmetic WISC-III Arithmetic WISC-III digit span, and spatial span subtests CANTAB Pattern memory and Spatial recognition memory test	Academic Achievement Executive Function Memory	Covariates: FSIQ
Crocker et al., (2009) USA	PAE *n* = 22, 36% F Age = 10.32(2.37) CON *n* = 20, 50% F Age = 9.96 (1.90) ADHD *n* = 23, 39% F Age = 9.78 (2.17) Range=6-13 .	Alcohol exposed (PAE) defined as 4+ drinks per occasion at least once a week, or 14 drinks per week on average during pregnancy.	Stated that "many of the children with PAE no longer reside with biological families" Statistics not provided.	PAE group primarily through multi-source collateral report Direct retrospective report by the caregivers of the CON and ADHD groups	Recruited as part of larger study of the "behavioural teratogenicity" of alcohol Professional referral, community outreach and self-referral. Nonexposed participants were recruited from the community via advertising at various agencies and child-related	Assessed by dysmorphologist for the facial features of FAS % of children meeting criteria for FAS not reported	VABS	Adaptive behaviour	Groups matched on age, sex, SES and race/ethinicity
Godel et al., (2000) Canada	PAE=19 CON=50 Mean age or range not reported, all participants grade 1 or 2 at time of data collection	PAE- less than five drinks more than once a week, or more than five drinks at any one time CON- none or moderate exposure	A drink referred to a bottle of beer, 120 ml wine, or a 30ml shot of spirits	Parent questionnaire Children excluded in alcohol intake information was vague, or not quantifiable	All children attending an elementary school, born between 1/6/1987 and 31/5/1989 eligible to participate	76% reported to display "criteria suggestive of FAS or FAE". Criteria not reported.	Pediatric Examination of Education Readiness Pediatric Early Elementary Examiniation ADD-H Comprehensive teachers Rating Scale (ACTeRS) Brigance Comprehensive Inventory of Basic Skills	Academic Achievement Attention Executive Functioning	Not stated Covariates None

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Family Characteristics	PAE source	Recruitment	FASD criteria assessed or diagnoses?	Measures/Test reported	Domains assessed	Notes
		(less than 5 drinks, less than once a week)					FAS/FAE Scale	FASD-specific behavior scale	
McGee, Bjorkquist, Price et al., (2009) USA	PAE *n* = 26, 46% F Age = 9.42(1.58) CON *n* = 26, 50% F Age = 10.03 (1.20) Range=7-12	PAE defined as 4+ drinks per occasion at least once a week, or 14 drinks per week on average during pregnancy	96.2% Con group in care of biological parents, 7.7% of PAE group in care of biological parents	PAE group primarily through multi-source collateral report	PAE recruited by professional or self referral CON recruited from community in child-related settings (child care centres)	Children assessed by dysmorphologist. *N*= 6 diagnosed with FAS, 18 without FAS, 2 missing. 23% FAS	Social Information Processing Interviews Test of Problem Solving	Social skills and social cognition Executive functioning	Covariates: Gender, Age
McGee, Bjorkquist, Riley et al., (2009) USA	PAE *n* = 25, 40% F Age = 4.47(0.86) CON *n* = 26, 46% F Age = 4.35 (0.85) Range = 3-5	PAE defined as 4+ drinks per occasion at least once a week, or 14 drinks per week on average during pregnancy.	Stated "many of the children with PAE no longer reside with biological families" Statistics not provided.	PAE group primarily through multi-source collateral report Con group via maternal report.	PAE recruited by professional or self referral CON recruited from community in child-related settings (child care centres). Matched on Age, Sex, Race, SES.	Children assessed by dysmorphologist and historical record review. 7 (28%) diagnosed with FAS.	CELF	Language	Covariates: None
Pearton et al., (2014) South Africa	PAE *n* = 15, 47% F Age = 7.05(0.50) CON *n* = 15, 53% F Age = 6.81(0.58) Range not reported	Included if any PAE. 5 participants from minimal, moderate and heavy consumptions recruited, but not subgroups analysis reported. Minimal < 5 Units/Week Moderate 5-10 units/Wk, or binge Heavy >10 units/wk or binge Unit= 10mL AA	26.7% of the PAE group in the care of a guardian	Semi-structured interview with parents or guardians.	Groups recruited from one of four schools in a rural municipality in South Africa.	Participants not assessed against FASD diagnostic criteria.	Test of Playfulness	Social skills	Covariates None
Rasmussen et al., (2011) Canada	PAE *n* = 37, 46% F Age = 5.1(1.6) CON *n* = 23, 9% F	PAE defined as caregiver report of "definitely yes" to	87% of CON group in care of biological parents	Retrospective report from primary caregiver (95% of ALC group in care	Both groups recruited from a respite program, eligibility criteria	Primary caregiver report of formal diagnosis of FASD.	SSIS	Social skills and social cognitiion	Covariates: Age

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Family Characteristics	PAE source	Recruitment	FASD criteria assessed or diagnoses?	Measures/Test reported	Domains assessed	Notes
	Age = 5.6 (1,8) Range = 3-8	PAE screening question. CON group defined as "definitely no".	5% of PAE group.	other than biological)	require SDQ total difficulty score in abnormal range.	60% had formal FASD diagnosis			
Vaurio et al (2008) USA	PAE *n* = 20, 60% F Age = 10.68(1.65) CON *n* = 20, 60% F Age = 10.36 (1.90) ADHD *n* = 20, 60% F Age = 9.52 (1.97) Range = 7-13	ALC defined as heavy exposure, at least 4 drinks per occasion at least once per week or 14 drinks per week during pregnancy	Not stated	PAE group primarily through multi-source collateral report Con group via maternal report.	ALC group recruited from clinic; professional- or self-referral CON group recruited through community postings	Children with PAE assessed by dysmorphologist and historical record review 10 (50%) diagnosed with FAS	WCST Controlled Word Association Test TMT	Executive function	"many of the children with PAE no longer reside with biological families" Statistics not provided. Covariates Age (COWAT test)
(Walthall, O'Connor, & Paley, 2008)	6-12 (*n*=130) PAE *n* = 100, 49% F Age = 8.59(1.6) CON *n* = 23, 9% F Age = 5.6 (1,8) Range = 3-8	Alcohol exposure assessing ed the "Health interview for Women" and groups dichotomised exposed or non-exposed	21% of the PAE group reported to be in the care of their biological mother 87% of the control group in the care of biological mother		Children with social skills deficits recruited through community posted flyers in health care providers, YMCAs and schools	All children assessed using 4-Digit diagnostic code 11% diagnosed with FAS, 43% with pFAS, and 46% with static encephalopathy, alcohol exposed	Mental Health Computerised Diagnostic Schedule for Children (4th Ed)	Affect regulation	Home placement controlled for in analysis predicting psychiatric symptoms.

Supplementary Table O.3

Characteristics of longitudinal studies comparing the cognitive and behavioural outcomes of groups of children exposed to various levels of alcohol

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Unit size	Cohort	Measures/Test reported	Domains assessed	Comments
Alati et al (2013) United Kingdom (UK)	N = 7062 Follow up at age 11	Never used, less than 1 glass per week, 1-6 per week, 1+ per day. Binge (4+ units AA/session) days over past month (never, 1-4, 5-10, 10+)	1 unit = 8gm AA	Avon Longitudinal Study of Parents and Children (ALSPAC)	National Curriculum Key Stage 2; UK benchmarks of education achievement in English, maths and science,	Academic Achievement	Binge = 4 drinks
Alvik et al (2013) Norway	N = 1,166 Follow up at age 5.5	Binge drinking in early pregnancy (during pregnancy weeks 0-6) ; no drinking, less than once per week, more than once per week	Standard unit not defined		Strengths and Difficulties Questionnaire (SDQ)	Affect regulation Executive Function	Binge = 5 to 7, or >8 drinks
Bay et al (2012) Denmark	N = 1,446 (analysis reported with n= 685) Follow up at age 5	Average drinks per week (0, 1-4, 5-8, 9+) during pregnancy	1 Std Drink= 12gm alcohol	Lifestyle During Pregnancy Study (LDPS). Prospective, follow-up study based on a sample from the Danish National Birth Cohort Study (DNBC)	Movement Assessment Battery for Children (MAB-C)	Motor Skills	Binge > 5 drinks Controlled for adverse experiences (See note above).
Falgren Eriksen et al (2012) Denmark	N = 1,629 Follow up at age 5	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+) Assessed during pregnancy (av. 17 weeks gestation).	1 drink = 12 gm alcohol	LDPS	Wechsler Primary and Preschool Scales of Intelligence (WPPSI)	Cognition	Binge = 5+ drinks
Furtado et al., 2016 Brazil	Total=56 ALC=28, 11.89 (0.42) CON=28, 12.00 (0.27) Range= 11-13,	Alcohol exposed defined as positive answer to any alcohol consumption questions in pregnancy, score above 1 on T-ACE, or positive ICD-10 diagnosis Not stated	Maternal report during pregnancy Any positive answer to alcohol consumption questions, a score or 1 or more on the T-ACE, or an ICD-10 diagnosis for harmful alcohol use	Recruited from women attending an obstetrical clinic,	N/A	D2 test of Attention WISC-III Verbal and performance scales RCFT Rey Auditory Verbal Learning Test (RAVLT) WCST Verbal fluency (categories and letters)	Attention Executive function Learning

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Unit size	Cohort	Measures/Test reported	Domains assessed	Comments
Humphriss et al., (2013) UK	N = 6,915 Follow up at age 10	None, Low (1-2 drinks/wk), moderate (3-7 drinks/wk), high (7+ drinks/wk). Binge; frequency over past month of 4+ drinks in one session. Reported in pregnancy	Drinks defined as "pub measure of spirits, half a pint of lager or cider, a wine glass of wine"	ALSPAC	Balance tasks	Motor Skills	Binge = 4+ drinks SES measured, unclear if home environment is assessed.
Kelly et al., 2013 UK	N = 9,597 Follow up at age 7	Light drinking (1-2 units per week) compared to no alcohol consumption. Those reporting more than light consumption were excluded	Unit defined as half a pint of beer, a glass of wine, or single measure of spirit or liquer	Millennium Nationally Representative longitudinal study	British Ability Scales Word reading assessment, and Pattern Construction assessment National Found for Educational Research Profress in Maths Test SDQ	Academic Achievement Executive functioning	Covariates included a summary of responses to questions about family context including discipline strategies, self-rated parenting competence, self-rated closeness with child, frequency of reading to child
Kesmodel et al.., (2012)	N = 1,628 Follow up at age 5	Abstainer, Light (1-4 drinks/week), Moderate (5-8), Heavy (9+) Binge drinking= dichotomised to any or none	12 grams	DNBC	WPPSI-R Behavioural Rating Inventory of Executive Functioning (BRIEF) Test of Everyday Attention (TEACH-5)	Attention Executive Functioning	Being = 5+ drinks See note above re: home environment in the DNBC/LDPS studies.
Korkman et al., (1998) Finland	Total=72 ALC= 46 CON = 26 Range= 5-9 FAS= 7 FAE= 13 (2 of 3 criteria; growth restriction, presence of dysmorphic features, CNS dysfunction)	Groups defined by alcohol exposure across different trimesters; 1- exposure during trimester 1, 2- exposure during trimester 1 and 2, 3- exposure throughout pregnancy, and non-exposed controls. Not stated	Included in PAE group if reported to use more than 10 drinks per week. Control group included in abstinence reported, and no pregnancy complications	Pregnant mothers reporting using moderate (10+ drinks per week), or found to have clinical or blood sample evidence of alcohol intoxication at prenatal visits to a tertiary maternity hospital were refer to the study.	FAS= 7 FAE= 13 (2 of 3 criteria; growth restriction, presence of dysmorphic features, CNS dysfunction) 15% FAS	Attention; inhibition and control, sustained concentration Receptive language; auditory analysis, repeating nonwords, the Token Test Naming; Naming Colours, Naming Body parts, Speeded Naming, Name Learning Visual-Motor; Visual motor precision, VMI Manual Praxis; Kinesethic praxis, dynamic praxis	Attention Motor Skills Memory Language

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Unit size	Cohort	Measures/Test reported	Domains assessed	Comments
						Verbal memory; word span, logical learning Visual memory; memory for faces.	
Niclasen, Andersen, Strandberg-Larsen, Teasdale., (2014) Denmark	N = 37,315) Follow up at age 7	No binge drinking, early binge drinking (binge drinking prior to 16 weeks gestation), late binge drinking (binge drinking in last 2 months pregnancy) Binge drinking defined as 5+ drinks on occasions	12 grams	DNBC	SDQ Mean scores on SDQ internalising (emotional and peer problems) and externalising (hyperactivity and conduct) Proportions above cut off scores on SDQ subscales; hyperactivity, conduct, emotional and peer problems	Affect regulation Executive Functioning	
Niclasen, Andersen, Teasdale, Strangberg-Larsen (2014)	N = 37,152 Follow up at age 7	Binge drinking frequency 0, 1, 2-3, 4+ binge episode in pregnancy, and cumulated total number of drinks Binge drinking defined as 5+ drinks on occassions	Not stated	DNBC	SDQ Mean scores on SDQ internalising (emotional and peer problems) and externalising (hyperactivity and conduct) Proportions above cut off scores on SDQ subscales; hyperactivity, conduct, emotional and peer problems	Affect regulation Executive Functioning	
O'Leary et al., (2010) Australia	N= 1,846 Follow up at age 2, 5 and 8.	None, low (<1 drink/day), moderate (1 drink/day), heavy (>1 drink/day).	10 grams	Randomly Ascertained Sample of Children born in Australia's Largest State (RASCALS)	CBCL	Affect regulation Attention Executive Functioning	Covariates included: Parental mental health Family functioning scale, Parenting Scale.
O'Leary et al., (2013) Australia	N= 4,680 Follow up at age 8	None, Low (1-2 drinks/session, <7/wk), moderate (3-4 drinks/session, =7/wk), occasional binge (5+ drinks/session, less than weekly, heavy (>7/wk, including binge drinking weekly or more frequently)	10 grams	RASCALS	Western Australian Literacy and Numeracy Assessment	Academic Achievement	

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Unit size	Cohort	Measures/Test reported	Domains assessed	Comments
Pesonen et al., 2009	N=289	Number of standard drinks (12g) per week during pregnancy, reported by mothers prior to leaving the maternity ward	12 grams	Finnish Birth Cohort	Sleep Disturbance	Sleep	SES and maternal education controlled for
Sayal et al., 2013	11 (KS2 *n*=10,558) (SDQ *n*=6,587)	None, >1 drink per week, <1 drink per week	8 grams	ALSPAC	SDQ Total problem sores (0-40) Conduct subscale Hyperactivity subscale [Emotional problems, and peer problems scales not reported]	Executive Function	Maltreatment proxies not reported.
Sayal et al., 2014	11 (KS2 *n*=6,939) (SDQ *n*=4,610)	At least one occasion of binge drinking (>4 drinks in a day)	8 grams	ALSPAC	SDQ Total problem sores (0-40) Conduct subscale Hyperactivity subscale [Emotional problems, and peer problems scales not reported]	Executive Function	
Sayal et al., (2009) UK	N=12,678 Follow up at ages 4 and 6.5	Binge (4+ drinks) per day in the second or third trimester	8 grams	ALSPAC	SDQ Total problem sores (0-40) Conduct subscale Hyperactivity subscale [Emotional problems, and peer problems scales not reported]	Affect regulation Executive function, hyperactivity, and impulsivity.	Note that two studies report different analyses of the same sample, albeit analysing either average alcohol consumption (Sayal et al., 2007) or binge drinking (Sayal et al., 2009).
Sayal et al., (2007) UK	N=12,678 Follow up at ages 4 and 6.5	Light (<1 drink/wk), moderate (>1drink/wk).	8 grams	ALSPAC	SDQ Total problem sores (0-40) Conduct subscale Hyperactivity subscale [Emotional problems, and peer problems scales not reported]	Affect regulation Executive function, hyperactivity, and impulsivity.	
Skogerbo et al., 2013	N = 1,617 Follow up at age 5	0, 1-5, 5+ drinks per week Number of binge episodes 0, 1,2, 3+	12 grams	DNBC	SDQ (SDQ reported as total, and prosocial)	Executive function Social Skills	Controlled for adverse experiences (See note above).
Sood et al., (2001) USA	N= 501 Follow up at ages 6-7	None, Low (<0.3 fl oz/day), heavy (>0.3 fl oz/day).	N/A; results expressed as fl oz of absolute alcohol.		CBCL	Affect regulation Executive function, hyperactivity, and impulsivity.	Includes an exposure to violence covariate, family structure and SES.
Underbjerg et al., (2012) Denmark	N= 1,628 Follow up at age 5	None, Light (<4 drinks/wk), moderate (5-8 drinks/wk), heavy (>9 drinks/wk).	12grams	LDPS	Test of Everyday Attention for Children (TEACH-5)	Attention	Small sample of heavy drinking cohort (n=20), wide confidence intervals Binge = 5+

Author, (Year) Country	Demographics Group= (N, mean age(SD)) Age range	Alcohol Exposure Definition	Unit size	Cohort	Measures/Test reported	Domains assessed	Comments
							Controlled for adverse experiences (See note above).

Supplementary Table O.4

Characteristics of studies examining alcohol exposure as a continuous variable

Author, (Year) Country	**Demographics** Group= (N, mean age(SD)) Age range	**Alcohol Exposure Definition**	**Unit size**	**Cohort**	**Measures/Test reported**	**Domains assessed**	**Comments**
Bailey et al., (2004)	5 (*n*=499)	Average alcohol consumption per day, and dichotomous yes/no report of binge drinking	Not defined	Detroit Sept 1989- Aug 1991 cohort	WPPSI TRF	Attention	Binge = 5 drinks Mainly African-American, low SES Children excluded if adopted out. "Community violence exposure included as covariate"
Burden, Jacobson, Sokol, Jacobson (2005) USA	N= 337 Follow up at age 7.5	Average alcohol consumption per day during pregnancy	Not defined	Detroit Sept 1986-Aug 1989 cohort	CPT WCST Tower of London WISC-III subtests (digit cancellation, coding, digit span, arithmetic), category fluency	Attention Executive functioning	Disruption of caregiving (separated from caregiver for at least 4 months included as co-variate)
Carter et al., (2016) ZA	ALC= 93 CON= 64 Follow up at ages 5 and 9	Average daily drinks during pregnancy Included if averaging >1.0 Oz AA, or >2 binge (5 drinks) occasions in first trimester. CON if <0.5 oz AA per day and no binge.	Not defined	South African Birth cohort	At age 5 JSAIS) At age 9 CVLT WISC-IV; Full Scale IQ, verbal comprehension index, perceptual reasoning index, working memory index, processing speed index D-KEFS letter and category fluency.	Cognition Executive Functioning Learning and Memory	
Chiodo, Da Costa, et al., (2010) USA	N= 462 Follow up at age 7	Average daily drinks during pregnancy	Not defined	Detroit Sept 1989-Aug 1991 cohort	Continuous Performance Test (CPT) Achenbach TRF	Attention	
Chiodo, Sokol, Delaney-Black, Janisse, & Hannigan, (2010)	N=75 Follow up at between 4 and 5	T-ACE scores	Not defined	Pregnant women presenting to the antenatal clinic of Wayne State University (Detroit, MI) between February 1999 and January 2001	WPPSI Object Memory CPT Divided Attention Pegboard Symbol Digit VMI Finger Tapping	Attention Memory Motor skills	

Author, (Year) Country	**Demographics** Group= (N, mean age(SD)) Age range	**Alcohol Exposure Definition**	**Unit size**	**Cohort**	**Measures/Test reported**	**Domains assessed**	**Comments**
D' Onofrio et al., (2007	N = 8,621 Ages 4-11	number of drinks per week, recalculated from 1/mo, 1/mo, 3-4 d/mo, 1-2 d/wk, 3-4 d/wk Nearly every day, Everyday [See D'Onofrio et al., 2007 for full conversion details].	Drink size not defined	National Longitudinal Survey of Youth	CBCL- Attention and Conduct Problems subscales	Attention Executive Function	
Eilertsen et al., 2017	N = 114,247 Follow up at age 5	AUDIT-C scores, based on first trimester drinking behaviour used as alcohol exposure measure. (Summed to a score for 0-12)	N/A	Norwegian Mother and Child Birth Cohort Study	Conner's Parent Rating Scale (CPRS-R) CBCL ADHD Sub-scale	Attention	
Goldschmidt et al (1996) USA	N= 522 Follow up at age 6	Average number of drinks per day during pregnancy	Drink size not defined	Magee Women's Hospital Cohort	WRAT-R reading, spelling and arithmetic subscales.	Academic Achievement	Excluded if mother lost custody, or child put up for adoption.
(O'Connor & Paley, 2006)	N=42 Age range= 4-5	Retrospective report of prenatal alcohol consumption Maximum number of drinks per drinking occasion	0.60 oz AA ~14grams	Recruitment from attendees of a university health clinic	Pictorial Depression Scale Family Interaction Puzzle Task; during which Child negative affect assessed as the degree of unhappiness, sadness and hurt expressed during a puzzle task	Affect Regulation	Excluded if the child had not been living with the mother since birth.
Richardson., (2002) USA	N= 593 Follow up at age 10	Average number of drinks per day, across each trimester. Binge drinking; frequency of 4+ drink/session.	Not stated	Magee Women's Hospital Cohort	WCST WRAML Trail-Making Test Stroop Test Grooved Pegboard CPT	Attention Executive function Memory Motor skills	Current caretaker interviewed if children not in care of biological parents (7%) Removal not considered a covariate.