Tailoring Chronic Pain Assessment to Children and Young People with Cerebral Palsy

Meredith Grace Smith

School of Allied Health Science and Practice

The University of Adelaide

A thesis submitted in total fulfilment of the requirements for the degree of

Doctor of Philosophy

March 2025

Table of Contents

List of tables iv
List of figures v
Publications included in this thesis vi
Other publications, presentations and awards arising from this thesis vii
Abstract x
Declaration xii
Acknowledgements xiii
List of abbreviations xiv
Chapter 1 - Introduction 1
1.1 Overview of the clinical problem of pain in cerebral palsy 1
1.2 Specific aims 4
1.3 Research questions 4
1.4 Thesis structure 5
1.5 Significance of this research 6
Chapter 2 – Literature Review 8
2.1 Cerebral palsy 8
2.2 Chronic pain 11
Chapter 3 – Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties (Study 1) 23
3.1 Preamble 23
3.2 Authorship declaration 23
3.3 Abstract 25
3.4 Background 27
3.5 Methods 29
3.6 Results 34
3.7 Discussion 42
3.8 Conclusion 48
Chapter 4 – Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification (Study 2) 49
4.1 Preamble 49
4.2 Authorship declaration 51

4.3 Abstract 53
4.4 Introduction 54
4.5 Methods 55
4.6 Results 60
4.7 Discussion 70
4.8 Conclusion 72
Chapter 5 – Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study (Study 3) 73
5.1 Preamble 73
5.2 Authorship declaration 74
5.3 Abstract 76
5.4 Introduction 77
5.5 Methods 79
5.6 Results 82
5.7 Discussion 98
5.8 Conclusion 102
Chapter 6 – The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy (Study 4) 103
6.1 Preamble 103
6.2 Authorship declaration 104
6.3 Abstract 107
6.4 Introduction 108
6.5 Methods 109
6.6 Results 117
6.7 Discussion 129
6.8 Conclusion 134
Chapter 7 – The Pain Interference Questionnaire and Fear of Pain Questionnaire for children and young people with cerebral palsy: examination of structural validity (Study 5) 135
7.1 Preamble 135
7.2 Authorship declaration 136
7.3 Abstract 138
7.4 Introduction 139
7.5 Materials and Methods 140

7.6 Results 145
7.7 Discussion 152
7.8 Conclusion 155
Chapter 8 – Discussion and Conclusions 156
8.1 Integrated summary of key findings 157
8.2 Clinical and research implications of thesis findings 160
8.3 Limitations 168
8.4 Future directions 170
8.5 Conclusions 173
Appendices 174
Appendix A: CP-adapted pain assessment tools 174
Appendix B: Supplementary material from Chapter 3 175
Appendix C: Supplementary material from Chapter 4 176
Appendix D: Supplementary material from Chapter 5 177
Appendix E: Supplementary material from Chapter 6 178
Appendix F: Supplementary material from Chapter 7 179
References 180
Talking Mats copyright statement 199

List of tables

Tables are numbered in order of appearance according to their chapter, with the first number indicating the chapter

Table 3-1 Inclusion and exclusion criteria for the search 31
Table 3-2 Hypotheses to evaluate construct validity and responsiveness 33
Table 3-3 Overall rating and quality of evidence for chronic pain interference tools for children and adults who are unable to self-report 40
Table 4-1 Study eligibility criteria 56
Table 4-2 Key topics and prompts in the semi structured interview guide..... 59
Table 4-3 Descriptive statistics of participants 61
Table 4-4 Qualitative data category and subcategory description..... 64
Table 5-1 Descriptive statistics of Delphi study participants 82
Table 5-2 Round 1 consensus results for FOPQ-C-SF modifications..... 84
Table 5-3 Round 1 consensus results for mBPI modifications 86
Table 5-4 Round 2 consensus results for FOPQ-C-SF modifications..... 91
Table 5-5: Round 2 consensus results for mBPI modifications 92
Table 5-6 Results from round 2 – consensus changes to the mBPI and FOPQ-C-SF 94
Table 5-7 Results from round 2: Discordant suggested changes to the mBPI and FOPQ-C-SF 96
Table 6-1 Strategies implemented to facilitate participation from young people with varying cognitive and communication abilities at different stages of the project..... 113
Table 6-2 Interview guide stages 114
Table 6-3 Stages of qualitative content analysis..... 116
Table 6-4 Participant demographics 117
Table 6-5 Qualitative coding schema with definitions..... 119
Table 6-6 Median duration completion times by tool and category 125
Table 6-7 Changes implemented after data review by author team and consumer advisory group..... 127
Table 7-1 Inclusion and exclusion criteria..... 141
Table 7-2 Participant demographics of participants who completed the PIQ-CP 146
Table 7-3 Participant demographics of participants who completed the FOPQ-CP.... 148
Table 7-4 Confirmatory Factor Analysis findings 150

List of figures

Figures are numbered in order of appearance according to their chapter, with the first number indicating the chapter

Figure 1-1 Thesis chapter outline 5
Figure 3-1 Graphical abstract for Chapter 3 26
Figure 4-1 Development of the survey instrument 57
Figure 4-2 Ratings of relevance, comprehensibility, clinical feasibility and the need for modification of pain coping tools 62
Figure 4-3 Ratings of relevance, comprehensibility, clinical feasibility and the need for modification of the multidimensional tools for chronic pain including assessment of 'impact of pain on emotional functioning' 63
Figure 4-4 Categories from the qualitative descriptive study. 65
Figure 6-1 Example of the first two items and response scale of the mBPI. 121
Figure 6-2 FOPQ-C-SF item 9 image ("Pain makes me think something bad will happen" 121
Figure 6-3 Boxplot of completion times of the adapted Fear of Pain Questionnaire Short Form (FOPQ) and Modified Brief Pain Inventory (mBPI), comparing differing communication needs, cognitive abilities and versions of the tools. 125
Figure 6-4 Example of a completed Talking Mat for the mBPI. 127
Figure 6-5 Example of the first two items and response scale of the FOPQ-C-SF. 129
Figure 7-1 Decision making tree to determine the most appropriate version of the tool to complete 144
Figure 7-2 CFA findings for the PIQ-CP 151
Figure 7-3 CFA results for the FOPQ-CP two factor structure 151

Publications included in this thesis

This thesis contains two published manuscripts, two manuscripts under review and one manuscript prepared for submission. There is no requisite for written permissions from the journal publishers as per the publisher guidelines. To ensure consistency, page numbers, tables and figures have been adjusted to conform to thesis formatting. All references are integrated into the final thesis reference list. Manuscript supplementary materials are included as appendices.

Publication 1 (Chapter 3)

> **Smith MG**, Farrar L.C., Gibson R.J., Russo R.N., Harvey A.R. Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties. Dev Med Child Neurol 2023; 65: 1029-42. https://doi.org/10.1111/dmcn.15535

Publication 2 (Chapter 4)

> **Smith, M.G**., Gibson, R.J., Russo, R.N. Karanicolas, S., Harvey, A.R. Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification. Qual Life Res 2024; 33: 2247–2259. https://doi.org/10.1007/s11136-024-03693-1

Publication 3 (Chapter 5)

> **Smith, M.G**., Gibson, R.J., Russo, R.N., Harvey, A.R. Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study. Pain Reports 2025. 10: e1304. https://doi.org/10.1097/pr9.0000000000001304

Publication 4 (Chapter 6)

> **Smith, M.G**., Gibson, R.J., Schibani, M., Russo, R.N., Thirumanickam, A., Harvey, A.R. The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy. Quality of Life Research (2025). https://doi.org/10.1007/s11136-025-03981-4

Publication 5 (Chapter 7)

> **Smith, M.G**., Gibson, R.J., Russo, R.N., Smith, N., Harvey, A.R. The modified Brief Pain Inventory and Fear of Pain Questionnaire for Children Short-Form adapted for cerebral palsy: examination of structural validity. *Developmental Medicine and Child Neurology, UNDER REVIEW*.

Other publications, presentations and awards arising from this thesis

Other publications during my candidature:

1. Harvey, A., Smith, N., **Smith, M.**, Ostojic, K., & Berryman, C. (2024). Chronic pain in children and young people with cerebral palsy: a narrative review of challenges, advances, and future directions. *BMC Med*, *22*(1), 238. https://doi.org/10.1186/s12916-024-03458-0

2. Smith, N. L., Gibson, N., Bear, N., Thornton, A. L., Imms, C., **Smith, M. G**., & Harvey, A. R. (2025). Measurement properties and feasibility of chronic pain assessment tools for use with children and young people with cerebral palsy. *Disability And Rehabilitation*, *47*(4), 840-854. https://doi.org/10.1080/09638288.2024.2362398

3. Smith, N. L., **Smith, M. G.,** Gibson, N., Imms, C., Thornton, A. l., & Harvey, A. R. (2023). Pain coping tools for children and young adults with a neurodevelopmental disability: A systematic review of measurement properties. *Dev Med Child Neurol*, *65*(3), 318-328. https://doi.org/https://doi.org/10.1111/dmcn.15410

Conference presentations & published abstracts during my candidature:

1. **Smith, M.G**., "Adaptation of two pain assessment tools for children and young people with cerebral palsy: a multi-stakeholder consensus study", oral presentation, Healthy Development Adelaide 20th Anniversary Celebration, Australia, Oct 23, 2024.

2. Smith, N., **Smith, M.G.,** Hogan, A., Doyle, C., Harvey, A.R., "Pain assessment in cerebral palsy: Exploring a biopsychosocial framework and novel assessment modifications to improve relevance and accessibility", 90 minute workshop, AusACPDM Cairns, Australia, Aug 1-3, 2024

3. **Smith, M.G.,** Harvey, A.R., Gibson, R.J., Schibani, M., Thirumanickam, A., Russo, R.N., "Development and testing of adapted versions of two chronic pain assessment tools for young people with cerebral palsy (CP)", oral presentation, AusACPDM, Cairns, Australia, Aug 1-3 2024

4. Ostojic, K., Mimmo, L., **Smith, M.,** "Working together in codesign health research: guidance on how, when, and why from people with lived experience, clinicians, and researchers.", 60 min seminar, AusAPCDM, Cairns, Australia Aug 1-3 2024.

5. **Smith, M.G.,** Smith, N. L., Thirumanickam, A., Harvey, A.R., "Pain assessment for children and young people with cerebral palsy: challenges and solutions", 90 minute workshop, Australian Pain Society, Darwin, Australia, 21-24 April 2024

6. **Smith, M.G.,** Harvey, A.R., Gibson, R.J., Karanicolas, SK., Russo, R.N., "Examining impact of pain on emotional wellbeing tools in children and young

people with cerebral palsy: stakeholder consensus for modification", oral presentation, PICH down-under webinar, 8th December 2023

7. **Smith, M.G**., Harvey, A.R., Gibson, R.J., Russo, R.N., "Tailoring chronic pain assessment to children and young people with cerebral palsy", oral presentation, PainAdelaide Conference, October 30 2023

8. **Smith, M**., Gibson, RJ. , Russo, RN., Karanicolas, S., Harvey, AR. (2023). (206.3) Modifying patient reported outcome measures to improve relevance and accessibility of chronic pain assessment for children and young people with cerebral palsy: a methodological approach. 30th Annual Conference of the International Society for Quality of Life Research. Quality of Life Research, 32(2), 23-220. https://doi.org/10.1007/s11136-023-03530-x

9. **Smith, M.G**., Harvey, A.R., Russo, R.N., Gibson, R.J., "Assessing the impact of chronic pain on emotional wellbeing in children and young people with cerebral palsy", poster presentation, International Symposium on Pediatric Pain, Halifax, Canada, October 1-4, 2023

10. Smith, N., **Smith, M.G.,** Hogan, A., Doyle, C., Harvey, A.R., "Co-designing pain assessment for children and young people with cerebral palsy", 90 minute hands on workshop, International Symposium on Pediatric Pain, Halifax, Canada, October 1-4, 2023

11. **Smith, M.G.** Farrar, L. C., Gibson, R. J., Russo, R. N., & Harvey, A. R., "Assessment tools for measuring chronic pain interference in children and adults who are unable to self-report: a systematic review of psychometric properties", poster presentation, Florey Postgraduate Research Conference, Adelaide, Australia, August 2, 2023

12. **Smith, M.G.** Farrar, L. C., Gibson, R. J., Russo, R. N., & Harvey, A. R., "Assessment tools for measuring chronic pain interference in children and adults who are unable to self-report: a systematic review of psychometric properties", Australian Pain Society Conference, April 1-4, 2023

<u>Invited & Funded</u>

1. Smith, N., **Smith, M.G**., Hogan, A., Doyle, C., Harvey, A.R., "Co-designing pain assessment for children and young people with cerebral palsy", 30 minute workshop, CP-Achieve symposium, Melbourne, Australia, Aug 3-4, 2023

2. **Smith, M.G**. & Smith, N., "Co-designing pain assessment for children and young people with cerebral palsy", rapid fire oral presentation, Pain in Child Health Knowledge Mobilisation Pain Education Day, Halifax, Canada, October 1, 2023

Awards during my candidature

2025 Australian Pain Society – Best Rapid Communication

2024 Healthy Development Adelaide – Best PhD Research Presentation

2024 Florey Postgraduate Conference – Innovation & Commercialisation (Clinical) Prize

2023 Healthy Development Adelaide – PhD Publication Award

2023 Florey Postgraduate Conference – Best Poster Prize (Allied Health)

Grants during my candidature

2025 Faculty of Health and Medical Sciences Early Grant Development Award
$19900

2024 Women's and Children's Health Network Early Career Spark Grant
$49000

2024 Adelaide Graduate Research School Travel Award
$3000

2024 Healthy Development Adelaide Travel Grant
$1000

2024 Faculty of Health and Medical Sciences Travel Grant University of Adelaide
$3055

2024 Faculty of Health and Medical Sciences Early Grant Development Award
$19915

2024 Allied Health Professional Travel Grant – Australian Pain Society Conference
$250

2023 PhD Travel Grant – Australian Pain Society conference
$500

2023 School of Allied Health Science and Practice Travel Grant
$1000

2023 School of Allied Health Research Development Support Grant
$14346

2023 Adelaide University Graduate Union/RC Heddle Travel Award
$4000

Abstract

Cerebral palsy (CP) is the most common childhood physical disability, affecting 1.6/1000 live births globally. Up to 75% of people with CP experience chronic pain, yet it is often poorly assessed and ineffectively managed. Best-practice pain interventions are underutilised in CP, partly due to a lack of valid and accessible assessment tools for those with diverse cognitive and communication abilities. This thesis identified, adapted and validated tools for assessing pain interference with function, including emotional functioning, in all children and young people with CP, regardless of cognitive and communication abilities.

A systematic review (Study 1) identified tools for assessing chronic pain interference in all children and adults unable to self-report. The Paediatric Pain Profile was recommended for children and adolescents due to its strong psychometric properties. Importantly, the review found many children and young people with diverse cognitive or communication abilities have the capacity to self-report but historically have lacked access to tools accommodating their needs.

To address this, young people with CP, parents and clinicians participated in a mixed methods study (Study 2) and Delphi study (Study 3). An online survey (n=61) identified chronic pain tools with the greatest potential for modification and validation for people with CP, from a list of tools deemed to have adequate psychometric properties and clinical feasibility. The modified Brief Pain Inventory (mBPI) and Fear of Pain Questionnaire for Children-Short Form (FOPQ-C-SF) rated the highest. A qualitative study (Study 2) (n=33) generated 58 tool modification suggestions, although some were conflicting. Subsequently, a modified electronic Delphi study (Study 3) (n=44) reached consensus on key modifications. Seven modifications were implemented for the FOPQ-C-SF and 21 for the mBPI, including visual presentation, wording and scoring changes. *Talking Mats®*, an evidence-based visual communication framework, was suggested to improve accessibility for those with diverse cognitive and communication abilities. Consequently, two administration methods were developed for each tool: standard pen/paper and *Talking Mats®*.

The comprehensibility and feasibility of the adapted FOPQ-C-SF and mBPI were established in a study of 22 children and young people with CP through modified cognitive interviews (Study 4), even for those with mild to moderate cognitive

impairment or complex communication needs. Studies 2-4 collectively provided sufficient evidence of content validity, the most important psychometric property according to the Consensus-based Standards for Measurement INstruments standards. The new tools were named the Fear of Pain Questionnaire for Cerebral Palsy (FOPQ-CP) and the Pain Interference Questionnaire for Cerebral Palsy (PIQ-CP).

Finally, Study 5 examined the structural validity of the FOPQ-CP and PIQ-CP to confirm suitability for research and clinical use. A cross-sectional study of children and young people with CP (n=128), who completed the adapted tools, found sufficient structural validity (CFI>0.96 or SRMR<0.09). Self-report was possible for 79% of participants, including 37% who would have required proxy-report without the *Talking Mats®* versions.

Overall, the FOPQ-CP and PIQ-CP are valid and feasible for young people with CP, including those with diverse cognitive and communication abilities. These tools can now be used in clinical practice to identify those who may benefit from tailored multidisciplinary pain interventions. The adaptation process used is appropriate for other tools and other populations with diverse needs.

Declaration

I certify that this work contains no material which has been accepted for the award of any other degree or diploma in my name in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. In addition, I certify that no part of this work will, in the future, be used in a submission in my name for any other degree or diploma in any university or other tertiary institution without the prior approval of the University of Adelaide and where applicable, any partner institution responsible for the joint award of this degree.

The author acknowledges that copyright of published works contained within this thesis resides with the copyright holder(s) of those works.

I give permission for the digital version of my thesis to be made available on the web, via the University's digital research repository, the Library Search and also through web search engines, unless permission has been granted by the University to restrict access for a period of time.

I acknowledge the support I have received for my research through the provision of an Australian Government Research Training Program Scholarship.

Meredith Grace Smith

March 2025

Acknowledgements

A heartfelt thankyou first and foremost to the children and young people with cerebral palsy, their parents and clinicians who so generously contributed to this project. Special thanks to the clinicians at Novita, particularly Kerry Evans, and the Women's and Children's Hospital for embracing this project.

Thank you to my primary supervisor, Professor Rachel Gibson, a remarkable leader, scientist and mentor. Thank you for your support, guidance, and kindness in creating opportunities for me to develop.

To my co-supervisors, Associate Professor Adrienne Harvey and Associate Professor Ray Russo. I never would have started this project without your encouragement! Adrienne, thank you for your unwavering support and vision in guiding this work. Thank you for your intentionality in pushing me forward for opportunities I would otherwise have not had. Ray, thank you for your wisdom and for exemplifying excellence and compassion in the care you provide for children and families.

To Linda, Amy and Caitlin, I am deeply grateful for your generosity and expertise in guiding this project. To Nadine Smith, and my colleagues at the School of Allied Health Science & Practice, thank you for sharing this PhD journey with me and for making it more fun along the way.

To Margo McKay, Eilidh Watson-Stanfield and Laura Holmes at *Talking Mats®* – thank you for your expert guidance, generosity and patience during the development of the *Talking Mat* versions of the pain assessment tools.

To my family, thank you for your encouragement throughout. To Adam, thank you for embracing this research with a level of enthusiasm uncommon among spouses of PhD candidates! I am thankful to God for the immense privilege of time and opportunity to complete this thesis.

List of abbreviations

AAC	Augmentative and Alternative Communication
AusACPDM	Australasian Academy for Cerebral Palsy and Developmental Medicine
BAPQ	Bath Adolescent Pain Questionnaire
BAQP-P	Bath Adolescent Pain Questionnaire – Parent report
BPI	Brief Pain Inventory
CALI	Child Activity Limitations Interview
CALI-P	Child Activity Limitations Interview-Parent
CAT	Computer Adaptive Test
CCN	Complex Communication Needs
CFA	Confirmatory Factor Analysis
CFCS	Communication Function Classification System
CFI	Comparative Fit Index
ClinROM	Clinician-Reported Outcome Measure
COREQ	Consolidated Criteria for Reporting Qualitative Research
COSMIN	COnsensus-based Standards for the selection of health Measurement INstruments
CP	Cerebral palsy
CPAQ	Chronic Pain Acceptance Questionnaire
CP-QOL	Cerebral Palsy Quality of Life scale
CSF	Custom Short Form
EDACS	Eating and Drinking Ability Classification System
EFFC	Effectiveness Framework of Functional Communication
EMR	Electronic Medical Record
ePROM	Electronic Patient-Reported Outcome Measure
FDI	Functional Disability Inventory
FOPQ-C	Fear of Pain Questionnaire for Children
FOPQ-CP	Fear of Pain Questionnaire for Cerebral Palsy
FOPQ-C-SF	Fear of Pain Questionnaire for Children - Short Form
FSF	Fixed Short Form
GCPS	Graded Chronic Pain Scale
GMFCS	Gross Motor Function Classification System
GMFM	Gross Motor Function Measure
GRADE	The Grading of Recommendations Assessment, Development and Evaluation
HREC	Human Research Ethics Committee

IASP	International Association for the Study of Pain
ICD-11	International Classification of Diseases
ICF	International Classification of Functioning, Disability and Health
IRT	Item Response Theory
MACS	Manual Ability Classification System
mBPI	Modified Brief Pain Inventory
NCCPC-R	Non-communicating Children's Pain Checklist-Revised
ObsROM	Observer-Reported Outcome Measure
PAICP	Pain Assessment Instrument for Cerebral Palsy
PBI	Pain Burden Inventory-Youth
PBI-C	Pain Burden Inventory-Caregiver
PCQ	Pain Coping Questionnaire
PCS-C	Pain Catastrophizing Scale for Children
PedIMMPACT	Pediatrics Initiative on Methods, Measurement and Pain Assessment in Clinical Trials
PedsQL	Pediatric Quality of Life Inventory
PES	Pain Evaluation Scale
PII-P	Pain Interference Index - Parent
PIPS	Psychological Inflexibility in Pain Scale
PIQ-CP	Pain Interference Questionnaire for Cerebral Palsy
PODD	Pragmatic Organisation of Dynamic Display
PPCI	Pediatric Pain Coping Inventory
PPP	Paediatric Pain Profile
PPQ	Varni/Thompson Pediatric Pain Questionnaire
PPST	Pediatric Pain Screening Tool
PREM	Patient-Reported Experience Measure
PRISMA	Preferred Reporting Items for Systematic reviews and Meta-Analysis statement
PROM	Patient-Reported Outcome Measure
PROMIS PPIS	Patient-Reported Outcome Measurement Information System Pediatric Pain Interference Scale
PROMIS PPPIS	Patient Reported Outcome Measurement Information System Pediatric Proxy Pain Interference Scale
PROSPERO	International Prospective Register of Systematic Reviews
PSOCQ-A	Pain Stages of Change Questionnaire for Adolescents
PVAQ	Pain Vigilance and Awareness Questionnaire

QPS	Questionnaire on Pain caused by Spasticity
r-FLACC	revised Face, Legs, Activity, Cry and Consolability scale
RMSEA	Root Mean Squared Error of Approximation
SRMR	Standardised Root Mean Squared Residual
VFCS	Visual Function Classification System
VSS	Viking Speech Scale
WHO	World Health Organization
WLSMV	Weighted Least Squares with Mean and Variance adjustment

Chapter 1 - Introduction

1.1 Overview of the clinical problem of pain in cerebral palsy

Cerebral palsy (CP) is the most common cause of childhood physical disability, with a global prevalence of 1.6/1000 live births.[1] CP is defined as 'a group of permanent disorders of movement and posture that are attributed to non-progressive disturbances occurring in the developing foetal or infant brain, causing activity limitation'.[2] Although CP is primarily a motor disorder, it is also associated with disturbances of sensation, perception, cognition, communication, behaviour, epilepsy and secondary musculoskeletal disorders.[2] Pain is one of the most common comorbidities associated with CP, with up to 76% of children and young people with CP reporting ongoing pain.[3] The prevalence of chronic pain in CP is higher than other paediatric populations[4], and adversely impacts quality of life[5], self-perception[5], psychosocial wellbeing[6], and participation in physical leisure activities[7]. Chronic pain is even more frequently reported in children with CP who have lower levels of gross motor function (i.e. non-ambulant), present with a dyskinetic motor type or who are of female sex.[3, 8] Although the prevalence of chronic pain in CP is well documented, the assessment and management of pain in CP has been lacking.[9]

A likely barrier to appropriate pain assessment in CP is the population heterogeneity, with varying severity of the motor disorder and its associated functional impact.[10-13] To group differing functional presentations of CP, classification systems are commonly used to describe gross motor[14], manual ability[15], communication[16], and eating/drinking[17] function. This wide-ranging spectrum of ability means that some children and young people with CP can communicate verbally, walk independently and have no cognitive impairment, whereas others may use augmentative and alternative communication (AAC), wheelchairs or mobility aids, and have mild to severe cognitive impairment, or any combination of the above. When assessing pain, the child or young person's communication, cognitive, motor and visual function impacts their ability to engage with existing self-report pain assessment tools. Self-report is considered the 'gold standard' for measuring pain in other paediatric populations, however few tools are currently considered accessible and feasible for children and young people with diverse abilities.[18] Interventional studies investigating pain management in CP have also been limited in quality by a lack of valid and reliable pain assessment tools for use

across the spectrum of ability seen in CP.[19] While there will always be a subset of the CP population for whom self-report is not possible due to severe cognitive impairment, it is likely, with modification, many of the children and young people with diverse communication, cognitive and functional abilities would be able to provide some form of self-report. Although many existing assessment tools have proxy-report options, it is imperative self-report is prioritised where possible to ensure accurate assessment of chronic pain.

Assessing chronic pain within a biopsychosocial framework is considered best practice, as it recognises pain as a subjective experience influenced by sensory, neurological, affective, behavioural and sociocultural factors.[20] While the exact contribution of each factor is unclear, this subjectivity emphasises the need for comprehensive assessment across all domains, rather than focusing solely on pain intensity.[20] The psychological and social contributors to the pain experience also highlight the deeply personal nature of pain, and therefore the need to self-report where possible.

In paediatric populations without disability, biopsychosocial frameworks for pain assessment are well established, with a range of appropriate assessment tools utilised covering domains such as pain interference with physical function, emotional function and sleep.[20] While several domains have been recommended for pain assessment in CP[21], it is not yet established which tools should be used. It is important chronic pain assessment in CP moves beyond pain intensity in order to better understand how pain interferes with function, including emotional functioning. This is crucial given the impact these areas have on long term pain outcomes and management recommendations.[22]

Assessing the two broader areas of pain interference and pain coping has been suggested as a starting point for young people with CP.[23] Kingsnorth et al. (2015) conducted a systematic review to identify assessment tools within these areas for potential use in young people with CP.[23] However, this research, along with a more recent systematic review[24], acknowledged many of the existing tools were not appropriate, or accessible, in their current form given the diverse cognitive, communication and motor abilities within the CP population. They were also not developed with consideration for a disability population, with some tools assessing activities which may not be relevant for a young person with CP, for example higher level motor abilities such as running or jumping.[22, 25] Consequently, many children and young

people with CP are missing out on the opportunity to self-report their pain, which leads to inequity in access to best practice pain interventions.[26] In order to better understand the impact of pain on all children and young people with CP, chronic pain assessment tools need to be better investigated to ensure they are relevant, accessible and feasible for use with children and young people with CP and with diverse abilities.

To ensure pain assessment tools are appropriately identified and rigorously evaluated, the gold-standard COnsensus-based Standards for Measurement INstruments (COSMIN) methodology is used throughout this PhD.[27-30] COSMIN recognise developing a new assessment tool is a time-consuming process and instead recommend, where possible, existing assessment tools are evaluated, investigated and adapted for the population of interest.[31] This methodology prioritises the sound development and content validity of the measure, comprising of the relevance of the tool to the construct and population of interest, the comprehensiveness of the tool in relation to the construct of interest and the comprehensibility of the tool for the target population. COSMIN consider the subsequent psychometric properties, such as reliability, construct validity and responsiveness to change, of secondary importance to ensuring the tool measures what it intends to and is relevant and understood by the target population. Given the importance of obtaining self-report from the individual wherever possible, this thesis considered three main groups of reporters within the population: 1) those who can self-report verbally or in written form with no adaptations to usual patient-reported outcome measure (PROM) administration methods, 2) those who require adaptations to self-report due to diverse communication, cognitive or motor abilities, and 3) those who are unable to self-report at all due to severe cognitive impairment.

Furthermore, this PhD project is informed by an advisory group of people with lived experience of CP to ensure the research is relevant, accessible and focused on the priorities of people with CP. This advisory group comprises of two young adults with CP and a parent of a child with CP and is representative of the diversity of cognitive, communication and motor ability seen in the CP population.

The overarching aim of this PhD project is to select, adapt and validate tools to assess chronic pain interference with function, including emotional functioning, in all children and young people with CP, regardless of cognitive and communication abilities. This

includes those who do not have the capacity to self-report at all (likely due to severe cognitive impairment) and those needing modification to enable self-report due to diverse abilities.

1.2 Specific aims

1. To critically evaluate the psychometric properties of tools that assess chronic pain interference in all children and young people who are unable to self-report
2. To identify and adapt key existing tools for the assessment of chronic pain interference with function, including emotional functioning, for children and young people with cerebral palsy
    a. To select and modify key tools for the assessment of chronic pain in children and young people with cerebral palsy
    b. To examine psychometric properties of the adapted chronic pain assessment tools in the cerebral palsy population

1.3 Research questions

1. What are the psychometric properties of chronic pain assessment tools used to assess chronic pain interference in children and young people who are unable to self-report?
2. What modifications/adaptations are required to key chronic pain assessment tools to most accurately represent children and young people with cerebral palsy?
3. What are the psychometric properties of the adapted chronic pain assessment tools for children and young people with cerebral palsy?

For this PhD thesis, "children and young people" refers to individuals aged 5-30 years. However, assessment tools available for individuals >2 years of age were considered as this is the age at which typically developing children develop the ability to verbalise and report simple pain language such as 'hurt' and 'ouch'.[32] The assessment of pain in neonates and infants with CP is a large and complex area outside of the scope of this PhD project. For further reading in this area, a recent review paper by Letzkus and colleagues (2021)[33] investigating assessments and interventions for chronic pain in young children with CP may be helpful. The cut off for young adulthood was also extended to 30 years on the advice of the consumer advisory group, recognising this

period may be extended for individuals with a disability due to delayed transitions to adulthood social roles.

1.4 Thesis structure

This thesis consists of eight chapters involving five studies. Chapter 3 and Chapter 4 are included as published manuscripts and Chapters 5-7 are included as manuscripts currently under review. The structure of the thesis is provided in Figure 1-1. This figure visualises the flow of the thesis, including the gaps in the evidence identified through the overall literature review in Chapter 2 and how this informed the selection of the systematic review topic and subsequent studies to modify existing pain assessment tools. The final discussion (Chapter 8) explores each of these studies and brings together the overall thesis findings.

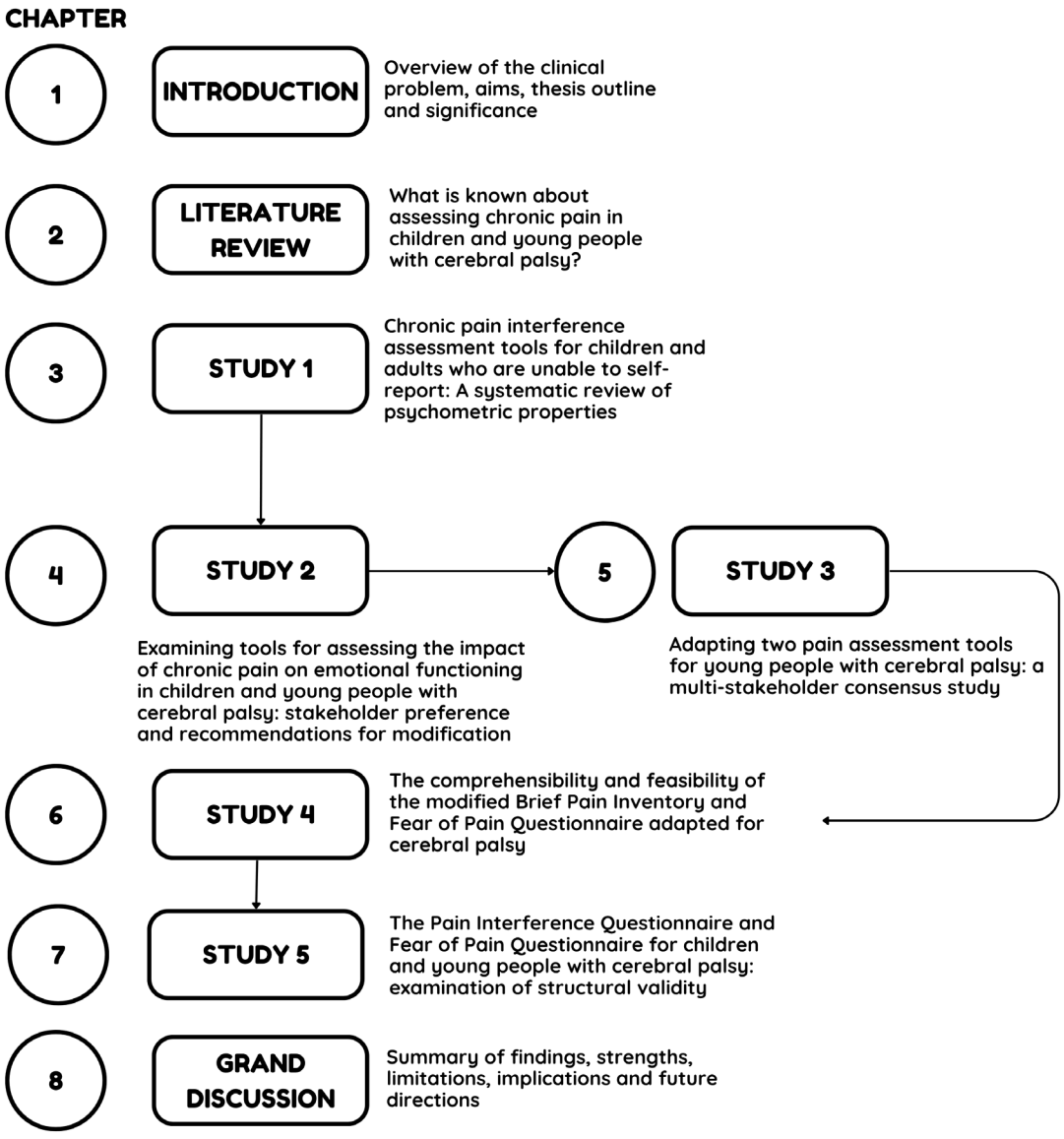


Figure 1-1 Thesis chapter outline

1.5 Significance of this research

Accurate assessment of chronic pain in all children and young people with CP, regardless of their ability level, is crucial in ensuring pain is appropriately managed. Best-practice interventions for chronic pain are currently under-utilised in CP, which is likely contributed to by inadequate assessment of pain.[26, 34] This PhD seeks to address an unmet need in identifying and adapting comprehensive pain assessment tools to be used across the spectrum of ability seen in CP, including enabling self-report and giving a voice to those who have been forgotten in the past. It is anticipated that improved assessment of pain, particularly for vulnerable groups of young people with CP and with diverse communication and cognitive abilities, will improve identification and referral to best practice multidisciplinary pain interventions. This, in turn, will improve pain outcomes for this vulnerable group, positively impacting quality of life.

The five studies in this thesis contribute new knowledge to improve assessment of chronic pain in all children and young people with cerebral palsy. The systematic review (Study 1) is the first study to identify and evaluate tools available for assessing how pain interferes with function in people who are unable to self-report at all. While other studies have recommended tools for identifying the presence of pain in people who cannot self-report, my thorough and critical review extends this by recommending the best assessment tools for understanding the functional impact of pain in this vulnerable group. The online survey in Study 2 fills a gap in knowledge by identifying which of the existing tools assessing impact of pain on emotional functioning should be further investigated and modified for children and young people with CP. The subsequent qualitative descriptive study identifies how these pain assessment tools should be modified for children with CP, with the Delphi study (Study 3) reaching consensus on modifications for implementation. While these recommendations were identified in the context of two specific assessment tools, many of the suggestions could be applied to outcome measures outside of the pain field to improve inclusive and accessible self-report options.

Studies 2 and 3 informed the development of modified versions of two pain assessments tools (modified Brief Pain Inventory and the Fear of Pain Questionnaire for Children), with Study 4 exploring the results of these tools being pilot tested in a group of children and young people with CP and with diverse communication and cognitive

abilities. The final study (Study 5) used these modified tools on a larger scale and provided robust evidence of validity.

This PhD project has involved modifying and testing two tools which will be made publicly and freely available for children and young people with CP to assess the impact of chronic pain, reducing the inequity previously seen in this population. These tools have been shown to be comprehensible and feasible for children and young people with diverse abilities and can now be used by clinicians and researchers to better understand the impact of pain on all children and young people with CP. Clinicians will be able to use these tools in clinical practice to more accurately identify and assess the functional impact of chronic pain, including being able to obtain self-report from some children and young people who would have otherwise relied on proxy-report alternatives. In research settings, there are opportunities for greater inclusion of children and young people with CP and diverse abilities in chronic pain intervention trials. Overall, this PhD has provided access to two valid and accessible chronic pain assessment tools for use across the population.

Chapter 2 – Literature Review

2.1 Cerebral palsy

2.1.1 Definition, prevalence & prognosis

Currently, the most recognised definition of CP was developed by Rosenbaum et al. (2007) through a series of workshops and panels.[2] It describes CP as a group of permanent disorders of movement and function resulting from a non-progressive injury to the developing brain. A key feature of this definition is the motor impairments are often linked to secondary comorbidities, including problems with perception, cognition, communication, behaviour, sensation, seizures and other musculoskeletal disorders.[2] This injury to the developing brain usually occurs before 2 years of age, and although the brain injury itself is non-progressive, it is widely recognised that secondary musculoskeletal issues can develop, progress and worsen over time with physical growth, muscle tone and ageing.[35] This definition was the first to specifically highlight the wide range of associated neurodevelopmental disorders and impairments, in addition to the movement disorder itself.[2] Recognising the associated disorders and impairments is crucial in understanding the heterogeneity within the CP population, with the functional impact of CP ranging from mild to severe.[1] Although CP is still the leading cause of childhood physical disability worldwide, both the prevalence and severity of CP diagnosis has decreased over the past 10 years in high income countries.[1]

A recent systematic review and meta-analysis suggests the global prevalence of CP is approximately 1.6/1000 live births.[1] In Australia, the current prevalence of CP is 1.5/1000 live births, which has decreased by 40% over the past 10 years.[36] CP can be acquired in the prenatal, perinatal or postnatal period, however the 40% decrease in CP diagnosis is primarily in the prenatally or perinatally acquired CP groups and thought to be associated with improved antenatal care.[1] The rate of post-neonatal CP has also declined over the past 10 years from 1.4 to 0.8 per 10000 live births; this decline has been less significant than the pre/perinatal groups.[36] Although post neonatally acquired CP only makes up approximately 6% of the cohort in Australia, these children experience proportionally more severe associated impairments such as blindness, deafness, non-verbal communication, moderate to severe intellectual impairment and

epilepsy.[36] While the severity of the associated impairments and motor disorder itself has an obvious impact on function, it also directly impacts the life expectancy of an individual with CP.[37]

Although the global life expectancy for adults with CP is similar to the general population, individuals with severe motor impairment, or those with severe cognitive disability, swallowing dysfunction, nutritional concerns or spinal deformities, have a shorter life expectancy regardless of geographical location.[37-39] This is primarily due to the impact on cardiovascular and respiratory function.[40] Of particular importance to this PhD thesis is the gross motor prognosis for a child with CP, given the known links between ambulatory ability and the presence of pain.[3, 41-43] The gross motor prognosis, or likely gross motor ability, of a child with CP can be indicated by their Gross Motor Function Classification System (GMFCS) level. Rosenbaum et al. (2002) conducted a longitudinal study of 657 children aged 1-3 years, representing the full spectrum of clinical severity of motor impairment in CP, to determine average ages and levels of gross motor skill achievement by GMFCS classification.[44] For a child classified as GMFCS level I, they will likely reach peak gross motor development by age 4-6, as assessed by the Gross Motor Function Measure (GMFM 66), whereas a child classified as GMFCS level V is likely to have reached peak gross motor development by 3 years of age. Gross motor development tends to plateau after the age of peak motor development[44, 45], however mobility may also decline into young adulthood.[46] Chronic pain can directly impact the loss of mobility into young adulthood, especially for children in GMFCS levels III-V, while also indirectly influencing other factors known to reduce mobility such as exercise, physical activity and participation.[7, 46, 47] Within CP, different motor types and functional classifications also impact the presence and severity of chronic pain.[8]

2.1.2 Classification and functional impact

CP can be classified in multiple ways, including by functional classification systems, distribution and motor type.[2] Predominant motor types are divided into three main groups: spasticity, dyskinesia and ataxia. Spasticity is a form of hypertonia in which resistance to passive movement increases with increased movement velocity.[48] Dyskinesia is a group term for hyperkinetic movements of dystonia and/or choreoathetosis, as it can be difficult to differentiate dystonia and choreoathetosis.[49] Dystonia presents primarily as involuntary movements that are either sustained or

intermittent, usually resulting in atypical postures or twisting movements.[48] Choreoathetosis differs from dystonia by featuring rapid, involuntary and jerky movements (chorea), or slower writhing movements (athetosis).[50] Ataxia presents as a disorder of balance and difficulty controlling the timing, force, rhythm and accuracy of coordinated movements.[50] Spasticity is the most common predominant motor type in CP, with Australian data suggesting 85% of children with CP have predominant spasticity.[51] This is followed by dyskinesia (7%), ataxia (4%) and hypotonia (3%), consistent with global data.[51, 52] Despite the fact that predominant dyskinesia only accounts for 7% of children with CP, it is increasingly recognised that many people with CP exhibit more than one motor type (e.g. both spasticity and dyskinesia).[50, 53] This is important as individuals with dyskinesia are more likely to experience chronic pain, and dyskinesia also directly affects functional impairment.[41] Both predominant motor type and functional impairment can impact the likelihood of experiencing pain, as well as impacting an individual's ability to report pain and therefore receive intervention.

In order to classify functional abilities in CP, the most commonly used systems are the GMFCS[14], Manual Ability Classification System (MACS)[15], Communication Function Classification System (CFCS)[16] and Eating and Drinking Ability Classification System (EDACS)[17]. Each classification provides a 5-level rating from I (least impaired) to V (most impaired). Strong evidence supports the psychometric properties of the GMFCS, MACS and CFCS in children with CP as descriptors, however these classification systems are not appropriate for detecting change.[54] Two newer functional classification systems, the Viking Speech Scale (VSS)[55] and Visual Function Classification System (VFCS)[56], are now being used more widely in clinical practice. The VFCS uses the same 5-level rating as the GMFCS, MACS and EDACS, whereas the VSS uses a 4-level rating; all scales rate from least to most impaired. The purpose of these scales is to broadly categorise the functional impact of CP, recognising the varying severity and complexity of physical disability and motor impairment for people with CP, with the condition becoming more complex with increasing number and severity of associated impairments.[57, 58] The severity of motor impairment in CP, along with the presence of comorbidities, places an individual at higher risk of pain.[3, 41-43] Despite this, all children and young people with CP are at higher risk of pain when compared to other paediatric populations.[3]

2.2 Chronic pain

2.2.1 Definition & incidence of chronic pain

The definition of pain was recently revised by the International Association for the Study of Pain (IASP) in 2020 as '*an unpleasant sensory and emotional experience associated with, or resembling that associated with, actual or potential tissue damage*'.[59] This updated definition recognises pain as a personal experience, influenced by biological, psychological and social factors, while clearly differentiating pain and nociception as two different phenomena.[59] Within this broader definition, it is also important to differentiate acute pain from chronic pain. Acute pain is recently occurring and of a shorter duration (usually less than one month),[60] with recent literature suggesting this time period could even be shortened to within the past seven days.[12] Acute pain is outside the scope of this thesis. However a good summary of acute pain in CP can be found in a prospective prevalence study by Ostojic and colleagues (2020).[12] Chronic pain is defined as '*persistent or recurrent pain lasting longer than 3 months, or longer than the expected time to heal'*. [61] This definition was developed by the International Classification of Diseases (ICD-11) and was adopted by the World Health Organisation (WHO) in 2019. Within the ICD-11, chronic pain has also been further classified into chronic primary pain, chronic cancer pain, chronic post-surgical and post-traumatic pain, chronic neuropathic pain, chronic headache and orofacial pain, chronic visceral pain and chronic musculoskeletal pain.[61] Pain is common in young people, with the newest figures finding one in five (20.8%) children and adolescents worldwide estimated to experience chronic pain.[62] The prevalence of chronic pain in the general paediatric population is higher in females and those with anxiety, depression and low self-esteem.[62, 63] Although chronic pain is common in the general paediatric population, the rates are even higher in children and young people with CP.[3, 4]

2.2.2 Chronic pain in cerebral palsy

2.2.2.1 Prevalence & aetiology

Children and young people with CP commonly report pain, with reported rates varying between 14-76% of the CP population.[3] For example, a Canadian study of 101 participants reported up to one third of children and young people with CP had moderate to high pain intensity trajectories, associated with worse physical and psychological wellbeing.[6] The variation in the reported pain prevalence across studies is likely due to heterogeneity in the studies themselves, including sampling bias in the

inclusion of age groups and motor types, and inconsistent measurement of pain.[3] Although chronic pain in young people with CP is reported across all GMFCS levels and motor types, it is even more common in those who also have severe motor impairment (e.g. GMFCS IV/V)[3, 41-43], are of older age[3], female sex[3, 64] and with dyskinesia[8]. The most commonly reported pain site across all GMFCS levels for people with CP is the foot and lower leg[65]. However those with predominant dyskinetic motor type are more likely to experience face, jaw and temple pain.[41] This is different to the general paediatric population, in which headache and general musculoskeletal pain are most common.[62]

Children and young people with CP experience the full spectrum of pain in a similar way to the general population. They also experience pain more specific to their disability.[4, 66, 67] Unique pain contributors in CP include hypertonia[13, 68], prolonged muscle contractures or sustained abnormal postures due to increased muscle tone, dyskinesia[41], hip subluxation/dislocation[69], joint contracture and scoliosis[13, 65, 70]. A cross-sectional study of 2736 children with CP in Sweden found the presence of postural asymmetries, associated with reduced ability to change position independently, doubles the likelihood of experiencing pain.[67, 71] Similarly, increased incidence of joint disease, particularly in the transition to adulthood, increases the likelihood of pain in CP.[72] Neuromuscular changes specific to CP, in conjunction with psychological factors and medical procedures experienced by people with CP, can contribute to an overall worsened chronic pain experience.[70] People with CP have also identified some specialised equipment used to manage their condition, such as ankle foot orthoses or wheelchairs, can contribute to their pain.[73, 74] Further condition-specific contributors to the overall chronic pain presentation include reduced physical activity, orthopaedic surgery, limited mobility and care-related procedures.[13, 74]

The type of pain experienced by a person with CP can also be variable and occur as the result of multiple mechanisms.[75] The classification of pain types in CP is an emerging and complex field and outside the scope of this PhD. For further reading on this complex area, consider recent work by Vinkel et al. (2021)[76] and Peterson et al. (2021)[75].

2.2.2.2 Impact & prognosis

Chronic pain affects children and young people with CP in many ways, including adversely impacting quality of life[5, 77], self-perception[5], psychosocial wellbeing[6],

sleep,[78] general activity[79, 80] and participation in physical leisure activities[7, 81]. The prevalence of chronic pain in CP is higher than other paediatric populations[4], and this trend continues into adulthood.[72, 80, 82, 83] As children with CP transition to adulthood, pain status does not tend to improve, with one study of 395 adults with CP reporting worse pain with increasing age.[84]

Even before entering adulthood, chronic pain does not appear to improve over time for young people with CP.[85, 86] In a study of 148 children with CP across all GMFCS levels, Christensen et al. (2017) found 45% had consistent pain, and 34% had worsening pain between routine clinical visits.[87] Consistent with these findings, a longitudinal study of pain in 67 adolescents with CP in Norway reported increasing pain prevalence, pain intensity, pain frequency, number of pain sites and interference with activities of daily living over a 5-year period.[88] Children in GMFCS levels III-V are less likely to report improvements in their pain status over time compared to those in levels I and II, which could be partly due to typically lower physical activity levels for those in GMFCS levels III-V.[87] The presence, and worsening severity, of chronic pain throughout childhood, adolescence and into adulthood has adverse impacts on many areas of life.

Chronic pain in CP is commonly associated with reduced physical activity levels[81, 89], participation[89], independence with activities of daily living[89] and quality of life[90], which is consistent with the general paediatric population.[63] Children with CP also specifically report adverse impacts on the ability to focus on schoolwork, participate in physical activity and sleep.[73] The impact of chronic pain on mental health in children and young people with CP should also not be underestimated.[42, 91-93] A study of 27 young people with CP found those with higher pain intensities were also more likely to use pain catastrophising as a coping strategy, despite catastrophising being associated with further increased pain intensity, pain interference, activity limitation, depression and worse psychological function.[79] Furthermore, as children with CP enter adulthood, it is likely chronic pain contributes to a loss of mobility and increases the risk of developing other chronic conditions.[7, 47] Adults with CP and chronic pain also have a 7-12% reduction in employment and a 2-8% reduction in earning if employed, compared to those with CP but without chronic pain.[94] The chain of events associated with unmanaged chronic pain as a comorbidity of CP, from childhood and into adulthood, increases the disease burden for individuals and their families, as well as potentially increasing the economic cost of the condition for the community.

2.2.2.3 Economic Cost

There is little research detailing the economic benefits of interventions in CP, including those for people with CP and chronic pain.[95] The Australian Physiotherapy Association (APA) delivered a report in 2020 on the economic value of physiotherapy.[96] The report did not specifically address chronic pain assessment and management in CP; however it determined physiotherapy for children with CP was overall cost effective.[95] Physiotherapy for children with CP provides a net benefit of $1502 per episode of care (estimated over a 12 month period), with a cost benefit ratio of 2.13:1 (Cost of $1326 with a $2828 quality of life value benefit).[95] The APA report acknowledged individuals with CP require ongoing intervention, and therefore these estimates are only valid for as long as intervention continues to be provided.[96] The APA report also reported the value of physiotherapy in managing chronic pain generally. The report focused on chronic neck pain, with results suggesting a cost benefit ratio of 2.72:1, demonstrating high cost-effectiveness compared to usual care.[96] It is therefore likely improving assessment and management of chronic pain in children with CP will not only improve quality of life for individuals but also deliver economic benefit.

2.2.3 Chronic pain assessment in paediatric populations

2.2.3.1 Chronic pain assessment in other paediatric populations

It is strongly recommended that chronic pain is holistically assessed within a biopsychosocial framework.[20] This framework recognises the complex interplay between biological, psychological and social factors in contributing to the experience of chronic pain, as well as highlighting how chronic pain can profoundly impact these same areas.[20]

In the general paediatric population, a core outcome set for paediatric chronic pain clinical trials was developed in 2008 by the Pediatrics Initiative on Methods, Measurement and Pain Assessment in Clinical Trials (PedIMMPACT) with core outcome domains of pain intensity, global judgement of satisfaction with treatment, symptoms and adverse events, physical functioning, emotional functioning, role functioning, sleep and economic factors.[97] Palermo and colleagues (2021) developed an updated core outcome set by following a systematic framework for selecting relevant outcome measures.[98] Consensus in this study was reached by collecting providers', patients' and parents' perspectives on the treatment of paediatric chronic pain, and inclusion of

mandatory and optional outcome domains. The core outcome set mandatory domains included pain severity, pain interference with daily living, overall wellbeing and adverse events.[98] The important, but optional, domains were emotional functioning, physical functioning and sleep.[98] The notable distinction between the PedIMMPACT domains[97] and the updated core outcome set by Palermo et al.[98] was the specification of 'pain interference with daily living' as a mandatory domain. The earlier PedIMMPACT domains recommended the Functional Disability Inventory (FDI) for assessing pain-related physical functioning in school-aged children (≥8 years of age), and the Pediatric Quality of Life Inventory (PedsQL) for children <8 years of age. Other recommended assessment tools to measure the impact of chronic pain on a child's function include the Bath Adolescent Pain Questionnaire (BAPQ) and the Patient-Reported Outcome Measurement Information System Pediatric Pain Interference Scale (PROMIS PPIS).[99, 100] The self-report versions of these tools are administered via a standard questionnaire (either electronic or pen/paper). Although the recommended tools have strong psychometric properties in the general paediatric chronic pain population, they have been less thoroughly investigated in CP.[23] Furthermore, before applying these assessment frameworks and tools to children and young adults with CP, it is important to consider whether there are domains of pain assessment that may be unique or more important to people with CP compared to the general paediatric population.

2.2.3.2 Assessment of chronic pain in the CP population

Chronic pain assessment and management has been identified as a priority area by people with lived experience of CP.[73, 101-104] Young people with CP have identified pain is often inadequately assessed and managed[105], with one study even suggesting clinicians tend to underestimate the prevalence of pain in the CP population.[13] Similarly to the general population, assessment of chronic pain within a biopsychosocial framework is strongly recommended in CP.[106-108] The most commonly used biopsychosocial assessment framework in CP is the International Classification of Functioning, Disability and Health (ICF), an internationally accepted model for the general assessment of children and young people with disability.[109, 110] The ICF includes five main components: body structure and function, activities, participation, environmental factors and personal factors. Each of these components are considered to provide an overall perspective of the person's health. Despite chronic pain in CP being strongly associated with lower quality of life and activity and participation

limitations[5, 77, 80, 81], many of the currently available pain assessment tools for children and young people with CP focus on body structure and function (e.g. pain intensity and severity) rather than the functional impact of pain.[11, 13, 106] Schiariti and colleagues mapped 31 pain assessment tools for children with CP to the ICF and found eleven included concepts of activities and participation, and only two included concepts of environmental factors.[106] Singular assessment of pain severity or intensity does not provide adequate insight into the activities or opportunities most important to the person, which are currently limited or impacted by chronic pain.[105, 111] However due to the complexity of assessment in the diverse and heterogenous CP population, there has been a tendency to focus on assessing pain intensity as the sole chronic pain outcome.[112] Unfortunately assessment of pain intensity alone does not appear to be sufficient to initiate adequate pain management.[19, 88]

Despite the recognised importance of pain assessment using a biopsychosocial framework, there was a clear gap in understanding which domains of chronic pain assessment were most important to people with CP.[21] To address this gap, Harvey et al. (2021) conducted a Delphi study with clinicians and people with lived experience to establish consensus on core domains for chronic pain assessment in children and young people with CP.[21] The twelve core domains identified were pain location, pain frequency, pain intensity, changeable factors, impact on emotional functioning, impact on participation, pain communication, influence on quality of life, physical impacts, sleep, pain duration and pain expression. These domains can be used to guide selection of pain assessment tools.

In contrast to the general paediatric population, few existing pain assessment tools are available to measure these domains. Of the assessment tools available, most are not validated in CP and few are accessible across the spectrum of cognitive, communication and motor ability seen in children and young people with CP.[23, 113] Some of the existing tools include items focussing on running or jumping, which may lack relevance to a child who uses a wheelchair for their mobility. Some tools are too long or complex to be used by an individual with a cognitive impairment, and others are difficult to engage with when using AAC devices. To appropriately assess chronic pain in CP, clinicians and researchers need to consider the heterogenous abilities of the population and how this might impact the ability to engage and meaningfully use existing assessment tools.

One of the first projects to consider the ability of an individual when making recommendations for pain assessment in CP was the Holland-Bloorview Pain Toolkit in 2015.[23] This toolkit summarised the results of a systematic review identifying chronic pain assessment tools for children with CP.[23] Tools were grouped based on whether they assessed how chronic pain interferes with daily activities (pain interference), or how an individual copes with chronic pain (pain coping). The tools identified for pain interference were the Bath Adolescent Pain Questionnaire (BAPQ), Body Diagram, Child Activity Limitations Interview (CALI), Non-Communicating Children's Pain Checklist-Revised (NCCPC-R), Paediatric Pain Interference Scale (PPIS), Paediatric Pain Profile (PPP) and the Varni/Thompson Pediatric Pain Questionnaire (PPQ). Although these tools had been validated in a paediatric population, only the NCCPC-R, PPIS, PPP and PPQ had been validated in CP.[23] A number of these tools were not suitable for all children with CP as they included items related to mobility, which were less relevant for GMFCS levels III-V.[23] Kingsnorth and colleagues (2015)[23] identified pain coping tools had a weaker evidence base pertaining to children with disabilities. However they were still able to identify eight available tools: Chronic Pain Acceptance Questionnaire (CPAQ), Graded Chronic Pain Scale (GCPS), Psychological Inflexibility in Pain Scale (PIPS), Pediatric Pain Coping Inventory (PPCI), Child Self Efficacy Scale (CSES), Pain Anxiety Symptom Scale (PASS-20), Pain Coping Questionnaire (PCQ), Pain Stages of Change Questionnaire for Adolescents (PSOCQ-A). Building on this work, a recent systematic review also identified no pain coping tools with content validity in neurodevelopmental disability, however identified three tools which show promise for use – CP Quality of Life (CP-QOL), Fear of Pain Questionnaire for Children (FOPQ-C) and BAPQ.[24]

More recent research has attempted to recommend pain tools for use in CP.[112, 114-116] This has been limited by a focus on pain intensity[112, 114], inadequate consumer involvement to ensure clinical feasibility[115], and a lack of robust psychometric property assessment of the tools[116]. Caravau et al. (2020) conducted '*Pain assessment in cerebral palsy: a systematic review of measurement properties and evaluation using the COSMIN checklist*', but did not differentiate acute pain from chronic pain.[114] The review identified fourteen instruments to assess pain in CP; the majority of these tools only assessing pain intensity. Further research on assessment tools for understanding the impact of pain was strongly recommended.[114] In 2023, Schiariti and colleagues updated the CP common data elements and did identify tools to capture the impact of

pain on function.[115] Although fifteen pain tools were identified and recommended, the study did not include consumer involvement to ensure tools would be suitable for end users, and no tools were identified to assess the impact of pain on emotional functioning, one of the core domains of pain assessment in CP.[21] Furthermore, psychometric properties were reported, but they were not evaluated using COSMIN, which is the current gold standard for development and evaluation of outcome measures in healthcare settings.[27] Noyek and colleagues (2024) completed a review of pain assessment tools for youth with brain-based disabilities and again highlighted the disparity in available outcome measures for domains other than pain intensity.[112] They also reiterated the difficulty of facilitating self-report for a population with diverse cognitive and communication abilities.[112]

One of the most significant challenges in assessing pain in CP is that some children and young people are unable to utilise self-report assessment tools in their current format due to cognitive and/or communication limitations.[13, 108] The Australian CP register estimates 38% of children with CP have some form of communication impairment at 5 years of age, with 24.5% considered non-verbal.[36] Furthermore, approximately 1 in 2 children with CP have an intellectual disability[57], with 21% of children with CP having moderate-severe cognitive impairment.[36] These statistics show there are likely complexities in assessing pain, particularly in enabling self-report, for almost half of the CP population. Particularly for children and young people with complex communication needs, it is often wrongly assumed they are unable to self-report, when in fact it is likely they could self-report if appropriate adjustments were made. Canada has recently released a *National Standard for Pediatric Pain Management*, which specifies the healthcare team is to '*provide appropriate support to the child and family to ensure effective communication that maximises the child's ability to self-report their pain experience*', including providing support for children with visual, hearing or communication impairment.[117] This is an important first step in recognising the importance of self-report in disability populations.

It is well accepted by the International Association for the Study of Pain that verbal description is only one of many indicators of pain.[59, 107] Vinkel et al. (2021) identified the specific challenge of diagnosing types of pain relying on subjective descriptions, specifically neuropathic pain and headaches, and recommended the development of new tools to assess these pain types in children and adults with limited ability to self-

report.[76] Several pain assessment tools have developed a proxy-report or observational component for use when a child is unable to self-report, including six of the pain interference tools identified by Kingsnorth et al. (2015) (BAPQ, CALI, NCCPC-R, PPP, PPIS, Varni-Thompson PPQ).[23] The PPP was also identified by Caravau et al. (2020), along with a further five proxy or observational tools primarily focused on pain intensity (Pain Evaluation Scale (PES), revised Face Legs Activity Cry and Consolability (r-FLACC), Child Facial Coding System (CFCS), Pain Assessment Instrument for Cerebral Palsy (PAICP) and Questionnaire on Pain caused by Spasticity (QPS)). [114]

Proxy reporting of pain is contentious as observers may not be sensitive enough to the pain experienced by the individual who is unable to self-report.[118] Breau et al. (2003)[119] found parents consistently underestimate their child's pain severity, whereas Ozcan et al. (2021)[120] reported no significant difference in the reporting of pain prevalence, intensity or the effect of pain on daily activities between mothers and adolescents with CP. It has also been suggested proxy reports completed by caregivers may be more closely related to the caregiver's own psychosocial distress, rather than the individual they are reporting for.[121] This incorrect estimation of pain is less concerning in the general paediatric population, as the proxy report can be compared to the child's own self-report. However for children and young people with CP and cognitive or communication impairment, self-report may not be possible or reliable.[118] There will always be some young people who, even with significant modification, will be unable to self-report due to severe cognitive impairment. Combining self-report, psychophysical methods of assessment (e.g. quantitative sensory testing), direct observation and caregiver report may provide the most reliable assessment of pain in these cases.[118]

For those people who can self-report using a method other than verbal or written communication, this should be prioritised and supported.[108, 122] This requires pain assessment tools that can be adapted and used with AAC systems.[112] Guard et al. (2021)[123] developed an iPad application (app) to assess the impact of chronic pain on individuals with developmental disabilities. This app aimed to address some of the concerns with other observational or proxy reported pain assessment tools by supporting self-report for children with cognitive impairment and complex communication needs. Given the recency of this app development, it has only been tested for face validity and user interface and is not yet available publicly; initial findings suggest healthcare professionals (primarily occupational therapists) may be able to use

the app for children with complex communication needs. The development study did not involve people with lived experience, and it is not clear how the items were selected, or the construct of interest defined.[123] We could find no other studies that have specifically sought to enable self-report of the impact of pain from children and young people with CP and diverse abilities.

Ensuring pain assessment tools are accessible for children and young people with CP and with diverse abilities is important, however it is also crucial these tools are robustly developed and examined to ensure they are valid, reliable and responsive to change in the CP population.[27] These three components of validity, reliability and responsiveness encompass several psychometric properties, which are indicators of the statistical strengths and weaknesses of an assessment tool.[27] Intervention studies, including systematic reviews of interventions for pain management in CP, have identified a lack of standardisation in the assessment of pain and a lack of validated outcome measures.[19] A lack of tools with strong psychometric properties limits assessment and evaluative ability across the recommended pain domains.[108] For example, an assessment tool with insufficient reliability cannot be used to accurately report on the outcomes of a pain intervention.

Without psychometrically robust and clinically accessible assessment tools for the spectrum of ability seen in CP, it is challenging to accurately identify the impact of pain on children and young people with CP or quantify change when undergoing chronic pain interventions. There is a clear research gap in understanding the clinical feasibility and measurement properties of tools for assessing the impact of chronic pain on individuals with CP.[23, 112, 114]

2.2.4 Improving pain assessment to improve pain outcomes

Understanding how to effectively manage pain in children and young people with CP is imperative. Both Ostojic et al. (2019) and McKinnon et al. (2020) identified a focus on pain management for children with CP and chronic pain was a priority for future research.[19, 101] There is a clear link between inadequate assessment of chronic pain and the difficulties faced in managing chronic pain in children and young people with CP.[9]

Assessing the impact of chronic pain in children and young people with CP who have historically been unable to self-report their pain, or access self-report measures, is essential in ensuring access to pain management. [124] Children and young people with

CP who cannot access existing self-report assessment tools, or clearly verbalise the impact of their pain, often have their pain underestimated or not recognised at all.[118, 119, 121] The absence of accurate pain assessment creates a significant barrier to managing pain effectively; if clinicians do not know pain exists, it cannot be treated appropriately. Furthermore, without valid and accessible assessment tools it is not possible to evaluate the outcomes of pain management interventions. Only by fully understanding the impact of pain, particularly in those historically under-assessed, is it possible to determine whether interventions should be pursued, and if they are effectively improving outcomes for children and young people with CP. Focusing on comprehensive pain assessment will likely also allow refinement of available interventions to ensure they are appropriately addressing the needs of individuals with CP. Although pain management is not the focus of this thesis, the improvement of pain assessment is likely to improve access to pain management interventions, and therefore enhance outcomes, for people with CP.

2.2.5 Conclusion

Chronic pain is highly prevalent in children and young people with CP and has significant negative impacts. This literature review has identified a clear gap in the availability of robust and accessible pain assessment tools for use across the diverse abilities seen in people with CP. Assessments that focus on the impact of pain, rather than pain intensity, are important in ensuring access to best practice pain interventions. While it is possible some existing pain assessment tools used in the general paediatric population may be useful for children and young people with CP, these will likely require adaptation and/or validation to enable self-report for those with diverse cognitive and communication abilities. Adapting these tools to improve accessibility will not be possible without the input of people with lived experience to guide, suggest and test changes made to the tools. It is also important to identify tools which can be used to assess the impact of pain in children and young people with CP who are unable to self-report at all, even with adaptations. Chronic pain assessment tools that are accessible, meaningful and valid for people with CP, and assess holistically within a biopsychosocial framework, are key to providing and evaluating treatment for chronic pain in CP. Improving the quality of pain assessment in CP has the potential to improve access to best practice pain interventions, which are currently under-utilised in CP, and

may lead to improved pain outcomes for children and young people with CP and chronic pain.

Chapter 3 – Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties (Study 1)

3.1 Preamble

Due to the paucity of evidence for assessing pain in children and young people with CP who are unable to self-report at all due to cognitive and/or communication limitations, my first study sought to identify tools for assessing chronic pain interference in all people who are unable to self-report. I could then determine if the identified tools may be appropriate for use in children and young people with CP who are unable to self-report. This systematic review was published in Developmental Medicine and Child Neurology. Links to supplementary materials are available in the appendices.

3.2 Authorship declaration

This manuscript is published in Developmental Medicine and Child Neurology

Smith MG[1], Farrar LC[1], Gibson RJ[1], Russo RN[2,3], Harvey AR[4,5]. Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties. *Dev Med Child Neurol*. 2023; 65: 1029–1042. https://doi.org/10.1111/dmcn.15535

[1]School of Allied Health Science and Practice, University of Adelaide, Adelaide, Australia

[2]College of Medicine and Public Health, Flinders University, Adelaide, Australia

[3]Paediatric Rehabilitation Department, Women's and Children's Hospital, Adelaide, Australia

[4]Neurodisability and Rehabilitation, Murdoch Children's Research Institute, Melbourne, Australia

[5]Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia

Statement of Authorship

Title of Paper	Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties
Publication Status	☒ Published ☐ Accepted for Publication ☐ Submitted for Publication ☐ Unpublished and Unsubmitted work written in manuscript style
Publication Details	Smith MG, Farrar LC, Gibson RJ, Russo RN, Harvey AR. Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties. Dev Med Child Neurol. 2023; 65: 1029–1042. https://doi.org/10.1111/dmcn.15535

Principal Author

Name of Principal Author (Candidate)	Meredith Smith		
Contribution to the Paper	Completed data collection, analysis of all data, wrote manuscript and acted as corresponding author		
Overall percentage (%)	65%		
Certification:	This paper reports on original research I conducted during the period of my Higher Degree by Research candidature and is not subject to any obligations or contractual agreements with a third party that would constrain its inclusion in this thesis. I am the primary author of this paper.		
Signature		Date	27/6/24

Co-Author Contributions

By signing the Statement of Authorship, each author certifies that:

i. the candidate's stated contribution to the publication is accurate (as detailed above);

ii. permission is granted for the candidate in include the publication in the thesis; and

iii. the sum of all co-author contributions is equal to 100% less the candidate's stated contribution.

Name of Co-Author	Lucy Farrar		
Contribution to the Paper	5% Contributed to data collection and analysis, edited manuscript		
Signature		Date	8/7/24
Name of Co-Author	Adrienne Harvey		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	27-06-2024
Name of Co-Author	Rachel Gibson		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	**27/6/24**
Name of Co-Author	Ray Russo		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	27/6/2024

3.3 Abstract

Aim: To identify and evaluate psychometric properties of assessment tools for assessing pain interference in children, adolescents, and adults with chronic pain and the inability to self-report.

Method: The protocol was registered with PROSPERO (CRD42022310102). A search was run in MEDLINE, Embase, and PsycInfo (29th March 2022) to identify articles reporting psychometric properties of pain interference assessment tools for children, adolescents, and adults with chronic pain and the inability to objectively self-report pain. Retrieved studies were reviewed by two authors (MGS, LCF) and study quality was assessed using COSMIN.

Results: Psychometric properties of 10 pain interference tools were assessed from 33 studies. The Paediatric Pain Profile (PPP) had low-quality evidence for content validity and internal consistency with children and adolescents who are unable to self-report. No tools for adults had evidence for content validity and internal consistency. No tool had evidence for all nine psychometric properties.

Interpretation: The PPP is recommended for pain interference assessment in children and adolescents with chronic pain and the inability to self-report. Few tools are available for adults. Three tools for children (Patient-Reported Outcome Measurement Information System Pediatric Proxy Pain Interference Scale; Bath Adolescent Pain Questionnaire for Parents; modified Brief Pain Inventory-Proxy [mBPI]) and three tools for adults (Doloplus-2; Patient-Reported Outcome Measurement Information System Pain Interference Scale-proxy; Brief Pain Inventory - Proxy) are promising but require further investigation.

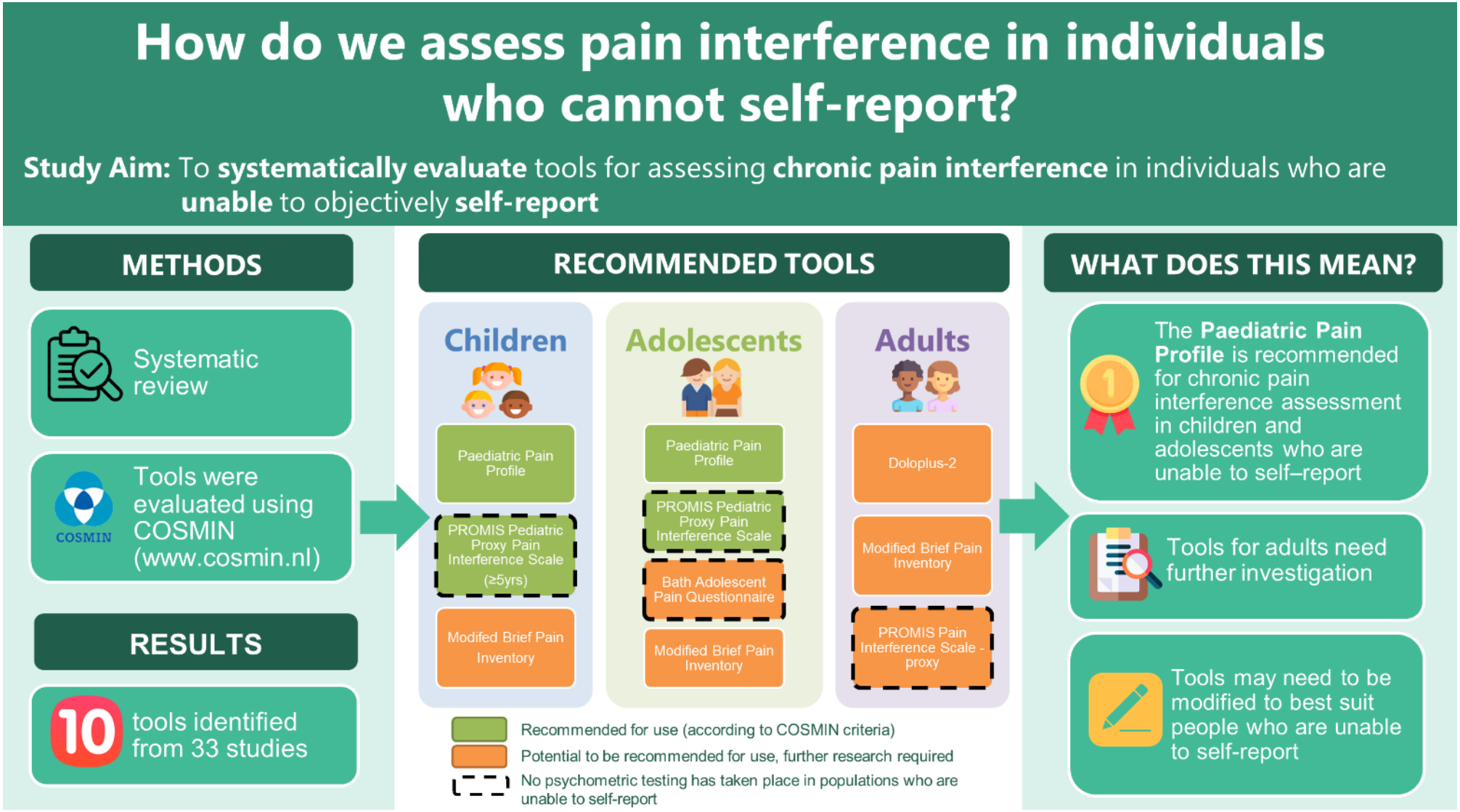


Figure 3-1 Graphical abstract for Chapter 3

3.4 Background

Chronic pain is a common condition worldwide, with prevalence estimated to be between 11% and 38% in children and adolescents and 11% to 20% in adults.[125-127] The prevalence of chronic pain is even higher in populations of individuals with disability, such as cerebral palsy, intellectual or developmental disability, and dementia, although pain assessment is often overlooked or misunderstood in this population.[3, 118, 128, 129] Despite the higher prevalence of pain, there are far fewer published studies on chronic pain management in these populations. A literature review by Barney et al. (2020) found only 33 published articles specific to pain in people with intellectual or developmental disability over a 5-year period, whereas for pain in people without intellectual or developmental disability there were more than 134 000 publications.[118]

Self-report is the criterion standard for assessing pain in children, adolescents, and adults. However, this can be challenging in populations where individuals may be unable to self-report owing to communication, motor, or cognitive limitations.[18, 118] It is also well accepted that verbal reporting of pain is only one of many indicators of pain.[59] For this review, 'unable to self-report' is the inability to self-report by any method, including verbal responses, written responses, and the use of assistive and augmentative communication devices. Individuals who are capable of self-reporting using a method other than verbal response should be supported to self-report where possible and are not the focus of this review. There are currently no clear recommendations on which pain assessment tools to use in clinical practice for individuals who are unable to self-report.[59, 118] Assessments covering functional dimensions of the pain experience are needed to capture information in a biopsychosocial model.[106] Core outcome sets have previously been developed for paediatric and adult chronic pain intervention trials, with 'pain interference with daily living' specified as a mandatory domain.[98, 130] The recommended measures to complete within these outcome sets rely primarily on self-report versions of patient-reported outcome measures, with the occasional parent-report tool included for paediatric populations.[97] Unfortunately, these options are insufficient when trying to assess pain interference in individuals of varying ages who are unable to self-report.

This systematic review focuses on pain interference assessment tools currently available for children, adolescents, and adults with chronic pain and the inability to self-report in any setting. As there is limited previous research in this area for individuals who are unable to self-report, this review aims to be inclusive and provide assessment recommendations across all age groups.[118] The inclusion of the adult population recognises that children and adolescents who are unable to self-report become adults who are unable to self-report, seeking to enhance continuity of care. Given the historical under-servicing of this population once they transition from paediatric services, it also seeks to provide greater access to pain assessment.[131] It is also possible that tools may be identified that have the potential to be used across age groups, or in future adapted for use in the adult population (and vice versa). This review will use the International Classification of Diseases, 11th Revision definition of chronic pain: 'persistent or recurrent pain lasting longer 3 months'.[61] Pain interference is defined by Karayannis et al. as 'a measure of the extent to which pain hinders engagement with physical, cognitive, emotional, and recreational activities, as well as sleep and enjoyment in life'.[132] For this review, pain interference is defined as referring to functional status, not symptom status or general health perception of overall quality of life, and does not include measures of pain coping, pain anxiety, or pain catastrophizing.[133] Tools included in this review will not be limited by which level of the International Classification of Functioning, Disability and Health (ICF) model they ascribe to, as long as they meet the definition of pain interference.[109]

Several systematic reviews have previously been completed on observational pain assessment tools for those with cognitive impairment[134-137], communication impairment[138], intellectual/developmental disability[23, 106, 114] and dementia[139-143]. However, these studies have primarily focused on acute and post-surgical pain, and on the domains of pain intensity, pain behaviour, and pain frequency. Pain intensity is an insufficient indicator of the impact of pain on an individual, limiting evaluation of management options.[144] Pain behaviour alone is also a challenging construct for individuals who are unable to self-report. Although pain behaviour may be appropriate for acute pain, it does not sufficiently capture chronic pain.[145] Many commonly observed pain behaviours also frequently occur for other reasons in individuals with disability, such as movements due to abnormal tone in those with cerebral palsy.[145] To our knowledge, there has not been a review completed on assessment tools for the

domain of pain interference in individuals with chronic pain and the inability to self-report.

The objectives of this systematic review were the following: (1) identify the available tools for assessing pain interference in children, adolescents, and adults with chronic pain and the inability to self-report; (2) determine the psychometric properties of the retrieved tools including validity, reliability, and responsiveness; (3) determine the feasibility and clinical utility of the retrieved tools for use in clinical practice.

3.5 Methods

The protocol for this systematic review was prospectively registered on PROSPERO (CRD42022310102) and the JBI systematic review database; it can be accessed online (at https://www.crd.york.ac.uk/prospero/display_record.php?RecordID=310102). The review was designed in accordance with the Preferred Reporting Items for Systematic reviews and Meta-Analysis statement (PRISMA) updated guidelines.[146] Two reviewers (MGS, LCF) completed title/abstract screening, full-text screening, data extraction, and quality assessment for all of the identified articles. They double-screened all the articles and both completed data extraction independently for all of the identified articles. Any disagreements between reviewers were resolved by discussion and a third author (ARH).

Search strategy

A comprehensive search strategy was designed by all authors and a medical librarian. An initial search was used to identify assessment tools that met the inclusion criteria, which also identified some articles on the psychometric properties of these tools for appraisal. A second subsequent search was then undertaken to ensure all articles assessing psychometric properties of the included tools were identified. Both searches included an adapted version of the translated COnsenus-based Standards for the selection of health Measurement INstruments (COSMIN) sensitive search filter for Ovid MEDLINE, developed to precisely retrieve articles on studies of psychometric properties.[147] The adapted filter was translated for Ovid Embase and PsycInfo. The first search was completed in MEDLINE, Embase, and PsycInfo from database inception to 15th February 2022. The second subsequent search was completed in the same databases, from database inception to 29th March 2022. Citation searching and targeted reference scanning were also used to minimize the likelihood of missing key

articles. A 'late breaking' search was completed on 29th July 2022 to identify any recently published, relevant articles. The individual search strategies for each stage are provided in Appendix B1. The electronic databases were searched by one author (MGS) and exported to an online systematic review management software, Covidence (Covidence systematic review software, Veritas Health Innovation, Melbourne, Australia; available at www.covidence.org).

Inclusion and exclusion criteria

The focus of this review is on individuals unable to self-report owing to limited expressive language, which may be the result of a communication, motor, or cognitive impairment. Emerson and Bursch suggest that the ability to self-report pain develops between 2 and 7 years of age.[32] The ability to self-report in this age range depends on the communication ability of the child, which can be variable for children of the same age, and the complexity of the assessment tool.[32] To ensure all potential tools were captured, tools were included if designed for individuals older than 2 years of age. Table 3-1 outlines detailed inclusion and exclusion criteria. Briefly, articles were included if they studied psychometric properties of an assessment tool that could be used to assess pain interference in children, adolescents, or adults with chronic pain and the inability to self-report. Exclusion criteria included assessment tools that (1) did not assess pain interference as a primary or secondary focus (one or fewer pain interference items, only assessed pain behaviour or assessed presence of pain during function rather than pain interference with function); (2) did not assess chronic pain (recall period < 24 hours); (3) were administered through self-report; or (4) were developed for assessing pain specific to a condition other than generalized chronic pain (e.g. headaches).

The psychometric properties of the tools were reported according to the criteria outlined in the COSMIN manual for evaluating the methodological quality of studies on measurement properties.[27-30] Studies that used the tool solely as an outcome measure in an intervention study were excluded.

Table 3-1 Inclusion and exclusion criteria for the search

Inclusion criteria	**Exclusion criteria**
Children, adolescents and adults (>2 years)	Children younger than 2 years where self-report is not developmentally appropriate
Experiencing chronic pain lasting longer than 3 months or longer than the expected time to heal	Pain identified in the article does not align with chronic pain as defined in this study (e.g. acute pain, procedural pain)
Assessment tool can be used by individuals who are unable to self-report	The tool is administered by self-report
Pain interference assessment measures containing a subscale that includes pain interference with daily living as a primary or secondary focus	Assessment tool did not measure pain interference as a primary or secondary focus
Study designs that investigate psychometric properties (could include validity, reliability, responsiveness and interpretability)	Psychometric properties were unreported for the pain assessment tool and/or the assessment tool was not validated Tool is used in an intervention study as an outcome measurement tool Systematic reviews on psychometric properties of assessment tools
	Other language translations of the included tools

<u>Study selection</u>

Titles and abstracts generated by both searches were screened against the eligibility criteria independently by two authors (MGS, LCF). The full text of the articles for all included titles/abstracts were obtained and screened against the eligibility criteria

again by the same two independent reviewers. Reference lists of selected articles were manually screened for additional articles meeting the eligibility criteria.

Data extraction and quality assessment

Data extraction was conducted independently by two reviewers (MGS, LCF) to minimize errors. Once completed, both reviewers reviewed data extraction together using a customized template in Covidence. Data were extracted and recorded under the following two headings. (1) Characteristics of the observer-reported outcome measure (ObsROM) or clinician-reported outcome measure (ClinROM): construct(s), target population, mode of administration, scales/subscales, number of items, response options, range of scores, time taken to complete, and available translations. (2) Characteristics of the studies examining psychometric properties: title, name of tool, psychometric properties assessed, population, and results. As there was significant variation in the terminology for different psychometric properties used by authors, the COSMIN definitions were used to determine the psychometric property assessed.[27] Where studies aimed to validate a single tool, we considered the psychometric property evidence of that singular tool. In studies that compared agreement between multiple tools, psychometric evidence for each of the eligible tools in this review was extracted. For the property of construct validity, hypotheses to test were developed (MGS, LCF, ARH) as per the COSMIN recommendations.[27] These hypotheses are listed in Table 3-2 and are based on the generic hypotheses developed by de Vet et al.[31]

Table 3-2 Hypotheses to evaluate construct validity and responsiveness

	Hypotheses
1	Correlations with changes in instruments measuring similar constructs (e.g. pain interference, functional disability) should be ≥0.50
2	Correlations with changes in instruments measuring related, but dissimilar constructs (e.g. pain intensity, pain catastrophizing) should be lower, i.e. 0.30–0.50
3	Correlations with changes in instruments measuring unrelated constructs should be <0.30
4	Correlations defined under 1, 2, and 3 should differ by a minimum of 0.10
5	Correlations between the self-report and proxy-report versions should be ≥0.70
6	Meaningful changes between relevant (sub)groups (e.g. patients with expected high vs low levels of the construct of interest)

Risk of bias was formally assessed using the 'COSMIN Risk of Bias tool to assess the methodological quality of studies on reliability or measurement error of outcome measurement instruments' and the 'COSMIN risk of bias checklist for Patient-Reported Outcome Measures (PROMs)'.[28, 29] As the focus of our review was on assessment tools for individuals unable to self-report, tools identified were primarily ObsROMs or ClinROMs.[28] Each study was assessed independently and quality assessment completed using the COSMIN risk of bias checklist (MGS, LCF). In addition, regular discussion occurred for all ratings until consensus was reached. For tools that had evidence of content validity in the included studies, two review authors (MGS, LCF) also completed an author rating of the content validity (comprehensiveness and relevance) of the overall tool as per the COSMIN methodology for evaluating the content validity.[30]

Study results for each tool and psychometric property were qualitatively pooled and each patient-reported outcome measure was summarized as sufficient (+), insufficient (−), inconsistent (+/−), or indeterminate (?) using the COSMIN criteria for good measurement properties. The overall content validity rating pooled results from the tool development study, content validity studies (if available), and the systematic review author rating. Briefly, each tool was assigned as follows: sufficient (at least 75% of results met the criteria); insufficient (at least 75% of results did not meet criteria);

inconsistent (individual study results were unable to be pooled and the inconsistency was unexplained); indeterminate (all study results were indeterminate). The modified Grading of Recommendations Assessment, Development and Evaluation (GRADE) approach was then used to rate the quality of the overall ratings as 'high', 'moderate', 'low', or 'very low'. Ratings were downgraded on the basis of indirectness, imprecision, risk of bias, and inconsistency, as detailed in the COSMIN guideline for systematic reviews.[27]

Recommendations were made for the most suitable assessment tool(s) for measuring pain interference in individuals of varying ages who are unable to self-report. These recommendations were based on overall evidence quality.[27] In accordance with COSMIN, the evidence for content validity and internal consistency, along with feasibility, were prioritized when determining recommendations.[27]

3.6 Results

Search results

The combined searches resulted in 2751 studies, of which 1976 were retrieved for title and abstract screening after duplicate removal. One hundred and sixty-six studies underwent full-text screening and 29 were deemed eligible for inclusion. A further four studies were identified and included through citation searching (Appendix B2). The final number of included studies was 33. These 33 studies (Appendix B3) reported on the psychometric properties of 10 pain interference assessment tools (Appendix B4), including nine ObsROMs[148-159] and one ClinROM[59]. Of the 10 tools assessed, two had short versions (Child Activity Limitations Interview-9[160] and Patient-Reported Outcomes Measurement Instrument System Pediatric Proxy Pain Interference Scale [PROMIS PPPIS] fixed short form[158]) and two had a computer adaptive test version (PROMIS PPPIS[158] and Patient-Reported Outcomes Measurement Instrument System Pain Interference Scale-proxy[159] [PROMIS PIS-proxy]). The Bath Adolescent Pain Questionnaire for Parents (BAPQ-P)[148] is a multidimensional tool, which means it contains a range of subscales each measuring a different domain of pain assessment, such as how an individual copes with pain or how pain interferes with their function. The BAPQ-P included three subscales relevant to this review: social functioning, physical functioning, and development. A summary of the excluded tools is provided in Appendix

B5. Twelve of the studies excluded at full-text review reported on psychometric properties of translated versions of the included tools: Doloplus-2 (Japanese[161], Chinese[162, 163], Norwegian[164-166], French[167, 168], German[169], Persian[170], Dutch[171]) and Paediatric Pain Profile (Brazilian Portuguese[172]. These articles are summarized in Appendix B6.

Study populations were broad and included children, adolescents, adults, and older adults with a variety of conditions including cerebral palsy[154, 155, 173, 174], burns[175, 176], traumatic brain injury[177], juvenile idiopathic arthritis[150, 178, 179], juvenile myositis[180, 181], stroke[182], musculoskeletal injury[183, 184], cancer[149, 157], cognitive impairment[153, 185] and idiopathic scoliosis[184]. Only five studies (3 tools) specifically investigated psychometric properties in the population of individuals who are unable to self-report.[153-155, 185, 186]The remaining 28 studies reported on a pain interference assessment tool administered by a method that could be used with individuals unable to self-report; however, psychometric properties were assessed in populations that could self-report. A summary of the pooled results for each tool and psychometric property, including author rating of content validity, is provided in Appendix B7. The risk of bias score for the psychometric properties in each individual study is reported in Appendix B8.

Tools for use with children and adolescents

BAPQ-P

The BAPQ-P was reported in three studies, with a pooled sample of 598 child/parent dyads as the same participant sample was used by Eccleston et al. and Cohen et al.[148, 187, 188] The BAPQ-P was developed for adolescents aged 11 to 18 years; however, the age range in the included studies was 5 to 18 years. The BAPQ-P content validity rating was assigned on the basis of a recent systematic review, in line with COSMIN recommendations to not re-rate content validity.[24] The BAPQ-P content validity study was tested in a population who could self-report. The other psychometric properties reported for this tool were internal consistency, reliability, and construct validity. Sufficient internal consistency was reported in two studies but rated as 'low' quality because there was no evidence of structural validity.[148, 187] Both test–retest reliability ('low' quality) and construct validity ('moderate' quality) were sufficient but downgraded owing to indirectness.[148, 187, 188] Reliability was further downgraded as only one study of adequate quality was available.[187]

Child Activity Limitations Interview-Parent

The Child Activity Limitations Interview-Parent (CALI-P) was developed for children aged 8 to 16 years. The psychometric properties of the CALI-P were reported in four studies, with an age range of 8 to 18 years.[150-152, 160] The overall content validity rating was of very low quality owing to an inadequate development study and no further content validity studies.[150, 151, 160] Structural validity was reported for both the 21- and 9-item versions of the CALI-P; however, results were inconsistent.[151, 160] Other psychometric properties reported for the CALI-P included internal consistency, reliability, construct validity, and responsiveness. The quality of all properties was downgraded owing to indirectness. Internal consistency was further downgraded to 'low' as it cannot be of higher quality than structural validity.[150-152, 160] Reliability was assessed as 'very low' owing to 'doubtful' and 'inadequate' quality studies.[150-152] Responsiveness was only reported in one study, resulting in a low-quality rating.[150]

PPP

The PPP is designed for children aged 1 to 18 years. Psychometric properties of the PPP were assessed in two studies of children aged 1 to 18 years.[155, 186] Content validity was assessed in one measure development study and was rated as 'low' owing to a lack of content validity studies.[155] The development study was completed through 46 interviews and a survey of 121 parents and caregivers of children with severe and life-limiting conditions who were unable to self-report. Other psychometric properties reported were internal consistency, test–retest reliability, construct validity, and responsiveness. All psychometric properties were assessed in children who were unable to self-report. Internal consistency was rated as 'low' owing to inconsistency and no reported structural validity.[155, 186] Two studies contributed to 'high' quality test–retest reliability and construct validity ratings.[155, 186] One study demonstrated 'moderate' quality responsiveness to change[186]

PROMIS PPPIS

The PROMIS PPPIS was designed for children aged 5 to 17 years. It had the highest number of relevant studies (*n* = 16), with ages of participants ranging from 2 to 18 years. Three studies demonstrated its content validity.[158, 173, 189] This was, however, subsequently downgraded to 'moderate' quality as content validity was not specifically

assessed for children unable to self-report.[158, 173, 189] Internal consistency was reported in one study but downgraded to 'low' owing to no reported structural validity.[184] As the PROMIS PPPIS is a direct translation of the self-report version, and the self-report version does have evidence for sufficient structural validity, it was not appropriate to further downgrade it to 'very low'.[190] Results were pooled for parent/child interrater reliability (Appendix B7); however, reliability was still rated as 'low' owing to inconsistent results and indirectness.[175, 176, 178, 189, 191] Construct validity was assessed in 11 studies, but downgraded to 'moderate' owing to indirectness.[174-180, 184, 192-194] When results were pooled, 76% of the hypotheses assessed were supported (Appendix B7) which met the 75% threshold for sufficient construct validity. 'Moderate' quality evidence for measurement invariance was found in the study conducted by Varni et al. who determined no significant difference between scores of children aged 5 to 7 years and those aged 8 to 17 years.[189] The PROMIS PPPIS psychometric properties were also analysed individually for the fixed short form, computer adaptive test, item bank, and custom short form versions (Appendix B9). Subgroup analysis of reliability and construct validity found 'very low' quality evidence of insufficient reliability and 'low' quality evidence of insufficient construct validity for the custom short form.[175, 176] The fixed short form had the strongest evidence of the four versions with 'very low' quality internal consistency[184], 'low' quality reliability,[178, 181, 184, 192, 193] and 'low' quality construct validity.[178, 180, 184, 192, 193]

Tools for use with children, adolescents, and adults

Modified Brief Pain Inventory

Only one study[154] investigated the psychometric properties of a proxy-report version of the modified Brief Pain Inventory (mBPI), described by Engel et al.[195] This study took place primarily in children with cerebral palsy who were unable to self-report (mean age 9 years); however, it included adults up to 34 years of age.[154] No content validity studies are available. The psychometric properties investigated were internal consistency, which had a 'low' quality rating owing to lack of reported structural validity, and construct validity, with 'high' quality evidence.[154] The mBPI is a promising tool that requires further investigation.

Pain Burden Inventory – Caregiver Report

The Pain Burden Inventory – Caregiver Report was designed for young people aged 7 to 21 years. It had no reported content validity studies. One study investigated its psychometric properties (mean age of participants was 14 years, age range not given), reporting internal consistency, child/parent interrater reliability, and construct validity.[156] This study was performed in a population of children who were able to self-report.[156] Internal consistency, reliability, and construct validity were all downgraded to 'low' owing to indirectness and imprecision. Internal consistency and reliability were further downgraded to 'very low' as the single study was of 'doubtful' quality.[156]

Pain Interference Index-Parent

The Pain Interference Index-Parent was reported in one study of children and young people aged 6 to 25 years.[157] It had 'low' quality content validity owing to a lack of studies and indirectness.[157] It was investigated in a population of children and young people who were able to self-report.[157] Internal consistency and construct validity were sufficient and indeterminate respectively; however, both were of 'very low' quality evidence. This was due to indirectness, imprecision, and the quality of the included study ('doubtful').[157]

Tools for use with the adult population

Brief Pain Inventory – Proxy Report

The BPI was reported in one study as a proxy-report tool in a study of adult patients with cancer, aged 31 to 88 years.[149] Participants were spouses, children of the patient, siblings, friends, or other relatives and were asked to 'answer as if they were the patient'.[149] The only psychometric property investigated was interrater reliability (patient vs proxy), which was found to be insufficient with 'low' quality evidence.[149]

Doloplus-2

The Doloplus-2 was investigated in two studies, with participants aged 65 to 96 years.[153, 185] The content validity rating was 'very low' owing to lack of content validity studies and inadequate measure development.[153] Structural validity was also 'very low' as only one study of inadequate quality was available and therefore internal consistency was also rated 'very low'.[185] Inter- and intrarater reliability was pooled (Appendix B7) and deemed sufficient and 'high' quality in two studies.[153, 185] Construct validity was

downgraded to ‘moderate’ quality as only one study of ‘adequate’ quality was available.[185] Cross-cultural validity was rated as ‘very low’ as the only available study was ‘inadequate’.[153]

PROMIS PIS-proxy

The PROMIS PIS-proxy had no reported content validity as it was developed as a self-report measure only for adults aged 18 years and older. Two studies reporting psychometric properties used the self-report measure as a proxy-report tool, with participants older than 62 years.[182, 183] ‘Moderate’ quality evidence for insufficient structural validity was reported in one study.[182] Both internal consistency and reliability were reported as indeterminate and of ‘low’ quality owing to indirectness and risk of bias, as the two studies were of ‘doubtful’ quality.[182, 183] Construct validity was reported; however, this was downgraded to ‘low’ owing to imprecision (small pooled sample size) and indirectness.[182, 183] Responsiveness was also downgraded to ‘very low’ owing to imprecision, indirectness, and risk of bias as only one study of adequate quality was available.[182]

Overall, 10 tools were identified (Table 3-3). Four were for use with children/adolescents, three with adults, and three that could be used across all populations. Only one tool, the PPP, had low-quality evidence for content validity and internal consistency in a population unable to self-report. It was designed for children and adolescents. None of the tools for the adult population had evidence for content validity and internal consistency. No tool had evidence for all nine psychometric properties.

Table 3-3 Overall rating and quality of evidence for chronic pain interference tools for children and adults who are unable to self-report

	Content Validity[1]				Structural validity[2]				Internal consistency[3]				Reliability/ Measurement error[4]				Construct validity[5]				Cross cultural validity/Measurement invariance[6]				Responsiveness[5]				**Overall rating[6]**
Outcome Measure	ROB	# Studies	Rating	Quality	ROB	#Studies	Rating	Quality	ROB	# Studies	Rating	Quality	ROB Score	# Studies	Rating	Quality	ROB Score	# Studies	Rating	Quality	ROB Score	# Studies	Rating	Quality	ROB Score	# Studies	Rating	Quality	
BAPQ-P	D	1	?	L					D	2	+	L	A	2	+	L	A	3	+	M									**B**
CALI-P	I	1	+	VL	A	2	?	L	V	4	+	L	D	3	?	VL	A	4	+	M					A	1	+	L	**B**
PPP	D	1	+	L					D	2	+	L	A	2	+	H	A	2	+	H					A	1	+	M	**A**
PROMIS – PPPIS	A	3	+	M					D	1	?	L	A	9	+	L	A	11	+	M	V	1	+	M					**A**
PROMIS - PIS					V	1	-	M	V	2	?	L	D	1	?	L	A	2	+	L					A	1	+	VL	**B**
Doloplus-2	I	1	+	VL	I	1	?	VL	V	1	+	VL	A	2	+	H	A	1	+	M	I	1	?	VL					**B**
mBPI									D	1	+	L					V	1	+	H									**B**
PBI-C									D	1	+	VL	D	1	?	VL	A	1	+	L									**B**
PII-P	D	1	+	L					D	1	+	VL					V	1	?	VL									**B**
BPI - proxy													V	1	-	L													**B**

Criteria for content validity rating

[a] Overall content validity is sufficient (+), insufficient (–), inconsistent (±), (indeterminant [?] rating not possible owing to availability of reviewers rating). +, The relevance rating is +, the comprehensiveness rating is +, and the comprehensibility rating is +. –, The relevance rating is –, the comprehensiveness rating is –, and the comprehensibility rating is –. ±, At least one of the ratings is + and at least one of the ratings is –. ?, Two or more of the ratings are rated ?.

Criteria for 'other measurement properties' rating

Measurement property rating: +, sufficient; –, insufficient; ?, indeterminate.

[b] +, CFA: CFI or TLI or comparable measure > 0.95 or RMSEA < 0.06 or SMRM < 0.08. ?, Not all information for + reported. −, Criteria for + not met.

[c] +, At least low evidence for structural validity and Cronbach's alphas ≥ 0.70 for each unidimensional scale of subscale. ?, Criteria for at least low evidence for structural validity not met. −, At least low evidence for structural validity and Cronbach's alphas < 0.70 for each unidimensional scale or subscale.

[d] +, Intraclass correlation coefficient ≥ 0.70. ?, Intraclass correlation coefficient not reported. −, Intraclass correlation coefficient < 0.70.

[e] +, The result is in accordance with the hypothesis. ?, No hypothesis defined by the review team. −, The result is not in accordance with the hypothesis.

[f] +, No important differences found between group factors (such as age, sex, language) in multiple group factor analysis or no important differential item functioning for group factors (McFadden's R^2 < 0.02). ?, No multiple group factor analysis or differential item functioning analysis performed. −, Important differences between group factors or differential item functioning were found.

[g] **Overall rating criteria**

A, Recommended for use and results obtained can be trusted. Assessment tools with evidence for sufficient content validity (any level) and at least low-quality evidence for sufficient internal consistency. B, Potential to be recommended for use, but they require further research to assess the quality. Assessment tools that do not fit criteria A or C. C, Not recommended for use. Assessment tools with high-quality evidence for an insufficient measurement property.

ROB score (COSMIN: COnsensus-based Standards for the selection of health Measurement Instruments)

V, very good; A, adequate; D, doubtful; I, inadequate.

Quality (modified GRADE: Grading of Recommendations Assessment, Development and Evaluation)

H, high; M, moderate; L, low; VL, very low.

Abbreviations: BAPQ-P, Bath Adolescent Pain Questionnaire for Parents; BPI-proxy, Brief Pain Inventory (used as proxy); CALI-P, Child Activity Limitations Interview-Parent; mBPI, Modified Brief Pain Inventory; PBI-C, Pain Burden Inventory-Caregiver Report; PBI-P, Pain Burden Inventory-Parent Report; PII-P, Pain Interference Index-Parent; PPP, Paediatric Pain Profile; PROMIS PIS, Patient-Reported Outcomes Measurement Information System Pain Interference Scale (used as proxy); PROMIS PPPIS, Patient-Reported Outcomes Measurement Information System Pediatric Proxy Pain Interference Scale; ROB, risk of bias

3.7 Discussion

This systematic review identified one tool, the PPP, with strong psychometric properties for assessing pain interference in children and adolescents (<18 years) who are unable to self-report. It is currently the only tool with sufficient quality evidence for content validity and internal consistency investigated in a population who cannot self-report.[155, 186] These are the two psychometric properties that COSMIN require to recommend a tool for use.[27, 196] The PROMIS PPPIS fixed short form is also recommended with a caution that psychometric testing has not specifically taken place in children older than 7 years who are unable to self-report. The mBPI is a promising tool that has established psychometric properties in children who are unable to self-report but requires further testing to provide evidence of content and structural validity.

There is lower-quality evidence for tools for use with the adult population. The tools available for the adult population should be used with caution as they require further investigation of their psychometric properties. The mBPI, although primarily tested in children, included an age range up to 34 years in one study, indicating this could also be an option for adults. The Doloplus-2 is promising for use in older adults but requires further evidence of content validity and internal consistency. Other tools that seem promising but require further content validity testing in populations that are unable to self-report are the BAPQ-P for adolescents (11–18 years) and the PROMIS PIS (as a proxy version) for adults.

All of the identified tools were feasible for use on the basis of availability, time required, and cost to complete (Appendix B4). Historically, pain intensity has been the outcome focus for pain management intervention. However, the way pain interferes with function is now understood as more important to individuals with chronic pain.[144] This shift in thinking probably explains why all of the tools identified for assessing pain interference were developed in the past 20 years.

In this review, we identified nine ObsROMs and one ClinROM for assessing pain interference in individuals who are unable to self-report. ObsROMs differ from ClinROMs as they are ‘observations made, appraised and recorded by a person other than the patient who does not require specialized professional training, e.g. proxy

measures'.[28] In line with COSMIN recommendations, the COSMIN risk of bias checklist for patient-reported outcome measures was applied to ObsROMs, and the risk of bias tool for ClinROMs was applied to ClinROMs.[28, 29] The ClinROM (Doloplus-2) was administered by health professionals (nurses) through observation only, whereas the PPP was administered by a combination of direct observation and proxy-parent report, with sufficient interrater reliability in health professionals and parents regardless of familiarity with the child.[155, 186] All of the other tools were administered by proxy-report only.

The accuracy of proxy-report was assessed in the included studies as either construct validity (correlations between proxy and patient scores) or interrater reliability (agreement between proxy and patient scores). These studies were conducted in populations of individuals who could self-report. Reliability results were, where possible, pooled for each tool (Appendix B7). COSMIN advise that, in the case of inconsistent results, studies only of 'adequate' and 'very good' quality can be used to determine the overall rating. As such, the pooled ranges for the PROMIS PPPIS parent/child interrater reliability did not include the results of Alcantara et al. as this study reported significantly different results and was rated as 'doubtful'.[27, 192] Although most of the tools demonstrated sufficient agreement (intraclass correlation coefficient > 0.70), it is unclear to what extent patient–proxy agreement is influenced by the ability to self-report pain. For example, an individual who can communicate with their proxy, either verbally or through assistive and augmentative communication devices, about how pain interferes with their function, may have a higher level of agreement than an individual who is completely reliant on their proxy to interpret the individual's pain. The PROMIS PPPIS reported higher agreement between parents and children when the parent or child themselves reported chronic pain.[194] This highlights the improved accuracy of the tool when chronic pain has already been identified and may be less useful in populations where pain is present but has not been identified.

The reliability of the proxy-report may be influenced by many factors. For example, higher levels of pain catastrophizing reported by caregivers using the PROMIS PPPIS resulted in overestimation of proxy-reported child pain interference.[191] The included studies most commonly used mothers or female caregivers as proxy reporters, but

some also included fathers, grandparents, siblings, children, household members, and friends. Unfortunately none of the studies compared the accuracy of different proxies. Further research to understand the relationship of personal, social, and cultural factors on a proxy's ability to accurately report pain interference is required.[118] Despite these factors, this review identifies proxy-report as one of only two available and feasible methods of assessing pain interference in individuals who are unable to self-report. We consider this is preferable to no assessment. It has been previously suggested that combining self-report, psychophysical methods of assessment (e.g. quantitative sensory testing), direct observation, and caregiver report may provide the most reliable identification of pain in people who are unable to self-report.[118] The PPP is the only tool identified in this review that uses a combination of approaches.

Although this review has focused on individuals who are unable to self-report by any method, it should be highlighted that, for individuals who do have some ability to self-report, self-reported communication should be supported and prioritized. Individuals should be supported to use their usual method of communication, whether this be 'high-tech' or 'low-tech'; however, a recent systematic review has suggested that lower-tech methods may be more feasible for use in real-world contexts.[122] This may involve adapting existing assessment tools to be more appropriate for use with augmentative and assistive communication devices; this can be done through strategies such as shortening the length of tools and adapting response options to be consistent with those available to the individual, either on their device or through other expression.

Only three of the included tools (PPP, Doloplus-2, and mBPI) were tested in populations of individuals who could not self-report. In addition, only two of these tools (PPP and Doloplus-2) had content validity in such populations.[153-155, 185, 186] This was a significant limitation in recommending tools for use in clinical practice. Content validity is considered the most important psychometric property by COSMIN, as it determines whether the tool measures the construct it claims to, and how relevant, comprehensive, and comprehensible the tool items are to the population of interest.[27] One parent-report tool (CALI-P) was developed without input from parents.[150] Two of the tools (mBPI and PBI-P) were direct translations of the self-report tools to proxy-report without re-testing content validity. A further two of the tools (PROMIS PIS and Brief Pain

Inventory-proxy) were not developed into a proxy-report form and instead asked the proxy to 'respond from the patient-proxy perspective'.[149, 182] The mBPI was modified by Engel et al. to be used as an outcome measure in neuromuscular disease without any robust content validity investigation, and then further modified for proxy-report with other psychometric properties investigated.[154, 195] COSMIN advises investigating the evidence for content validity of a tool first, followed by structural validity/internal consistency, then investigation of all other psychometric properties.[27, 30] This aims to reduce wasting resources on investigating the psychometric properties of a tool without sufficient content validity.

Only one of the tools identified (PROMIS PPPIS) investigated measurement invariance by comparing the proxy reports of children aged 5 to 7 years with those aged 8 to 16 years.[189] Establishing content validity for people who are unable to self-report is important; however, it can be challenging if those individuals are unable to participate in qualitative research. This is particularly difficult in the adult population where individuals may not have a consistent caregiver. An alternative method of assessing the psychometric properties of translated proxy-report tools is to investigate measurement invariance in different populations.[31, 197] This may be achieved by comparing groups of individuals with the same condition and severity level, but differing abilities to self-report.

The Doloplus-2 and the PPP were the only tools identified in this review with information on test–retest reliability (intra- and interrater reliability) and measurement error.[153, 155, 185, 186] Reliability is considered an essential psychometric property by COSMIN, but should be investigated after sufficient content validity and internal structure of the tool are obtained.[27, 31] The most important reliability measure for clinical practice is test–retest reliability and measurement error. The remaining studies assessed interrater reliability between the self-report and proxy-report versions only. This poses a potential limitation to interpretation of scores in clinical practice, as the smallest detectable change score is not available. Finally, none of the included articles assessed reliability or responsiveness to change of the tools over more than two time points. Further investigation of this would be beneficial in understanding the usefulness of these tools in longitudinal studies.

All of the tools identified had good face validity and feasibility. The utility of the tools, however, could be affected by the degree of other impairments an individual who is unable to self-report may also have. For an individual with reduced functional mobility as well as the inability to self-report, a tool such as the PROMIS PPPIS or PROMIS PIS-proxy will probably be difficult to use as many of the items relate to higher-level mobility tasks. Given the frequent co-occurrence of cognitive impairment and significant physical disability, clinicians and researchers need to consider the relevance of the tool items to the population they are assessing.

This review has highlighted the lack of chronic pain interference assessment tools for adults who are unable to self-report. Only 3 of the 10 tools were specifically for use in the adult population, with a further three having been designed for children/young people but also used in younger adults (up to 34 years of age). The only tool that was investigated specifically in an adult population who could not self-report was the Doloplus-2. The mBPI was also tested in a population who could not self-report, primarily in children, but included adults up to 34 years of age. This may be due to a lack of focus on pain management for older adults with cognitive impairment, or the historical under-servicing of those with neurodevelopmental disability once they transition to adult services.[131] Several of the excluded tools assessed pain behaviour or pain intensity in adults who were unable to self-report, but only in acute settings such as intensive or postoperative care. It is important that adults with the inability to self-report are given the same opportunity to express how pain interferes with their function and to access evidence-based chronic pain management. The lack of available tools may also highlight the difficulty obtaining proxy-report when many adults do not have consistent caregivers in the same way children do.

This study had several strengths and limitations. A strength of this review was the use of COSMIN, the recommended standard for assessing the measurement properties of outcome measurement instruments.[196] The review also focused on a specific area of chronic pain assessment which, to our knowledge, has not been reviewed, in a vulnerable group with historically under-reported pain.[118]

A potential limitation was the decision to exclude studies reporting psychometric properties of other language versions of the included tools. It should be noted that no tool was identified that was solely in a language other than English; all the tools that were reported in other languages had a translated version in English (Appendix B4, Appendix B6). In addition, relevant studies in any language were included. This decision was made as COSMIN advise new translations of an outcome measurement instrument should have psychometric properties investigated independently from other language versions, with content validity of primary importance when recommending tools for use.[31] The quality assessment process for COSMIN content validity involves a subjective rating (by the systematic review authors) of the relevance and comprehensiveness of the assessment tool itself, including rating item wording and response options (Appendix B7). Each different language version of an assessment tool contains linguistic or cultural nuances that cannot be accurately assessed by someone who is not fluent in that particular language. As none of the review authors were fluent in a language other than English, it was not deemed appropriate to provide a content validity rating of the other language translations of the included tools. This is a limitation; however, it is preferable to providing an inaccurate rating of content validity (and therefore inaccurate recommendations for use) of other language translations of the included tools. Overall, the available data and summarized results for each included tool was comprehensive and not limited by this exclusion.

Several commonly used tools were excluded from this review as they did not meet the eligibility criteria for 'pain interference'. Tools such as the Functional Disability Inventory[198] and the World Health Organization Disability Assessment Schedule[199], which assess overall health/well-being or disability, were excluded as they were not specific to pain-related functional disability. We determined a recall period of at least 24 hours was appropriate to assess the construct of chronic pain, acknowledging it is difficult for individuals to recall greater periods of time (e.g. >3 months as per the definition of chronic pain). For this reason, the Non-communicating Children's Pain Checklist-Revised, although previously used in chronic pain studies, was not included as its recall period is within the past 2 hours.[200] The exclusion of these tools was not considered to be a significant limitation. The included tools can be used to specifically

assess the interference of chronic pain on an individual's life, which is an important and under-reported aspect of pain assessment.[144]

Future research should focus on demonstrating further evidence for psychometric properties of assessment tools that could be used with people who are unable to self-report and with chronic pain, particularly the adult population. Evidence for content validity should be prioritized, which may require tools to be modified to better represent the specific population or condition of interest. Recommendations for a combination of pain interference assessment tools that use different methods such as direct observation, proxy-report, and physical testing could be valuable for people who are unable to self-report. Research using ethnographic approaches, or innovative technology, to investigate pain communication in people who are unable to self-report may provide greater insight into the individual's pain experience, making progress towards self-determination for this population.[201] Finally, examination of the strengths and limitations of ClinROMs compared with ObsROMs for people who are unable to self-report may also provide guidance for clinicians and researchers when selecting assessment tools.

3.8 Conclusion

This review has identified 10 tools for assessing chronic pain interference in children and adults who are unable to self-report. The PPP is recommended for use in children and adolescents (<18 years). No recommendation can be made for tools for use in adults without further psychometric testing

Chapter 4 – Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification (Study 2)

4.1 Preamble

Tools to assess chronic pain in children and young people with cerebral palsy; which ones to use?

In my systematic review in Chapter 3, I recommended the Paediatric Pain Profile (PPP) for assessing chronic pain interference in children and adolescents who are unable to self-report.[202] The PPP is a robust and clinically feasible tool which covers nine of the twelve core domains for pain assessment in CP (pain frequency, pain intensity, pain duration, pain expression, changeable factors, pain communication, impact of pain on emotional functioning, impact of pain on physical abilities, impact of pain on sleep).[113] The PPP was developed for children with CP with severe neurological impairment, and therefore is an appropriate tool for use in children and adolescents with CP, chronic pain and the inability to self-report. Having identified a high-quality tool for assessing chronic pain interference in children and young people with CP who are unable to self-report, I decided to focus the rest of this thesis on tools for those who can self-report but may need adaptations to do so.

When considering assessment of chronic pain in CP, it is essential to ensure full assessment of all the core domains. Available and accessible tools for assessing the impact of pain on emotional functioning are especially limited.[24, 113] For the purpose of this thesis, impact of pain on emotional functioning is broadly defined as 'the mind's response to living with pain'[203] and includes constructs such as pain-related fear, anxiety, low mood, and depression, pain catastrophising and pain coping. Emotional functioning status has been identified as the most important health and functioning outcome to children with CP and their parents, while also playing a key role in predicting the use of positive pain coping strategies.[26, 111] Factors contributing to the impact of pain on emotional functioning, such as pain coping and pain catastrophising, influence

pain-related physical disability, anxiety and depression [204, 205] and are also key targets for pain-related psychological interventions.[206] For example, a recent systematic review found no tools assessing pain coping could be recommended for use in the CP population, primarily due to a lack of evidence for content validity.[24] This is concerning as although positive pain coping strategies are recognised as an important self-management intervention for chronic pain[207, 208], individuals with CP utilise fewer positive pain coping strategies than other paediatric populations.[26, 34, 209] In order to improve access to best practice pain interventions, the impact of pain on emotional functioning needs to be accurately assessed in children and young people with CP. Given the lack of assessment tools available in this domain, the decision was made to focus the subsequent studies within my thesis on tools assessing the impact of pain on emotional functioning.

Following the publication of my systematic review in Chapter 3, Smith N et al. (2024)[113] identified **all** available tools for chronic pain assessment in CP, which included tools from the systematic reviews mentioned above[23, 24, 112, 114, 115] and my systematic review[210]. Each of the available tools were assessed for clinical feasibility and psychometric robustness using the COnsensus-based Standards for Measurement INstruments (COSMIN).[211] Few tools were identified with robust psychometric properties in the CP population, and those that could be recommended primarily assessed pain intensity. While assessment of pain intensity is important, how pain interferes with function is understood as more important to individuals with chronic pain.[112, 144] Smith N and colleagues again highlighted the particular need for tools to assess the impact of pain on emotional functioning for children and young people with CP.[113] They also emphasised the need to consider the feasibility of using existing tools across the range of communication, cognitive and functional abilities seen in the population. For example, the PROMIS Pediatric Pain Interference Scale (PPIS) has been previously recommended for use in CP as a pain interference measure[116], despite it being difficult to use in children with greater mobility limitation due to the inclusion of higher-level mobility tasks.[212] This highlights the importance of including representation across the spectrum of CP when determining appropriate assessment tools for practice.

A further challenge in ensuring feasibility is determining how best to support children and young people with CP and complex communication needs to self-report where possible.[112, 113] As discussed in Chapter 3, several assessment tools provide a parent/proxy report alternative, however given the inconsistency of proxy report these should only be used when the individual has no capacity to self-report on the construct of interest.[118] In all other circumstances, self-reported communication should be prioritised, which may require tools to be modified.[209]

Understanding which tools are meaningful and feasible for assessing the impact of pain on emotional functioning requires input from all key stakeholders, including people with lived experience of CP and health professionals. My next study was designed to identify the most appropriate tools for assessing the impact of pain on emotional functioning in CP, along with recommendations to modify these tools to be more appropriate and accessible. This process, which included individuals with CP, parents of children with CP and health professionals, also contributed evidence for the content validity of the tools in CP, a key factor in recommending tools for use in clinical practice and research.[24, 29] The results of this study were published in *Quality of Life Research*. Supplementary materials related to this article are provided as appendices.

4.2 Authorship declaration

This manuscript is published in Quality of Life Research

Smith, M.G.[1], Gibson, R.J.[1], Russo, R.N.[2,3], Karanicolas, S.[4], Harvey, A.R[5,6]. Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification. *Qual Life Res*. 2024; **33**: 2247–2259. https://doi.org/10.1007/s11136-024-03693-1

[1]School of Allied Health Science and Practice, University of Adelaide, Adelaide, Australia

[2]College of Medicine and Public Health, Flinders University, Adelaide, Australia

[3]Paediatric Rehabilitation Department, Women's and Children's Hospital, Adelaide, Australia

[4]School of Dentistry, University of Adelaide, Adelaide, Australia

[5]Neurodisability and Rehabilitation, Murdoch Children's Research Institute, Melbourne, Australia

[6]Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia

Statement of Authorship

Title of Paper	Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification
Publication Status	☒ Published ☐ Accepted for Publication ☐ Submitted for Publication ☐ Unpublished and Unsubmitted work written in manuscript style
Publication Details	Smith, M.G., Gibson, R.J., Russo, R.N., Karanicolas, S., Harvey, A.R. Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification. *Qual Life Res* (2024). https://doi.org/10.1007/s11136-024-03693-1

Principal Author

Name of Principal Author (Candidate)	Meredith Smith		
Contribution to the Paper	Completed data collection, analysis of all data, wrote manuscript and acted as corresponding author		
Overall percentage (%)	65%		
Certification:	This paper reports on original research I conducted during the period of my Higher Degree by Research candidature and is not subject to any obligations or contractual agreements with a third party that would constrain its inclusion in this thesis. I am the primary author of this paper.		
Signature		Date	27/6/24

Co-Author Contributions

By signing the Statement of Authorship, each author certifies that:

i. the candidate's stated contribution to the publication is accurate (as detailed above);

ii. permission is granted for the candidate in include the publication in the thesis; and

iii. the sum of all co-author contributions is equal to 100% less the candidate's stated contribution.

Name of Co-Author	Sophie Karanicolas		
Contribution to the Paper	5% Contributed to data collection and analysis, edited manuscript		
Signature		Date	2/7/24
Name of Co-Author	Adrienne Harvey		
Contribution to the Paper	10% Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	27-06-2024
Name of Co-Author	Rachel Gibson		
Contribution to the Paper	10% Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	27/6/24
Name of Co-Author	Ray Russo		
Contribution to the Paper	10% Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	27/6/2024

4.3 Abstract

Purpose: To firstly identify tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy (CP), and secondly identify suggestions to improve their relevance, comprehensiveness, comprehensibility and feasibility for the CP population. Improving assessment of the impact of pain on emotional functioning can enhance quality of life by improving access to interventions for pain-related physical disability, anxiety and depression.

Methods: Ethics approval was granted through the Women's and Children's Health Network Human Research Ethics Committee (2022/HRE00154). A mixed methods study with people with lived experience and clinicians, and guided by the Consensus-based Standards for Measurement Instruments (COSMIN), was undertaken. An online survey identified the highest rated tools for validation and/or modification for young people with CP and chronic pain. Focus groups and interviews investigated content validity and feasibility of the tools identified as highest rated.

Results: The Fear of Pain Questionnaire for Children-SF (FOPQ-C-SF) and Modified Brief Pain Inventory (mBPI) were the highest rated for pain coping and multidimensional assessment (respectively) from the online survey (n=61) of eight tools presented. Focus group and interview data (n=30), including 58 unique modification suggestions, were coded to six categories: accessibility, comprehensibility, feasibility, relevance, presentation and comprehensiveness.

Conclusion: Potential modifications have been identified to improve the appropriateness and feasibility of the FOPQ-C-SF and mBPI for children and young people with CP. Future research should implement and test these modifications, prioritising the involvement of people with lived experience to ensure their needs are met alongside clinicians.

4.4 Introduction

Cerebral palsy (CP) is the leading cause of childhood disability, with a global prevalence of 1.6/1000 live births.[1] Young people with CP experience chronic pain more commonly than other populations, with rates between 14% and 76% of the population.[3] Despite this, chronic pain in CP remains poorly understood and managed [12].

Inadequate chronic pain assessment in CP may stem from underutilisation of assessment tools.[13] Harvey et al. (2021) established consensus on twelve core domains for chronic pain assessment in CP [21], however few validated tools exist.[23] Furthermore, available tools lack suitability across the varying cognitive, communication and motor abilities in CP.[24, 210] Recent reviews highlight a particular need for valid tools assessing impact of pain on emotional functioning, particularly pain coping.[23, 24] Pain coping includes three key psychological factors (pain anxiety, pain catastrophising and fear of pain) which influence pain-related physical disability, anxiety and depression, all with significant impact on a person's quality of life.[204, 205] These factors are also targets for psychological interventions, influencing outcomes of the domain, 'impact of pain on emotional functioning'.[206] Multidimensional tools assessing impact of pain on emotional functioning may assist in identifying individuals who would benefit from further assessment of pain coping, and subsequently improve access to currently underutilised pain coping interventions.[34, 100]

Content validity is an essential measurement property of any tool; ensuring a tool measures what it claims to measure. This includes relevance to the intended population and construct, comprehensiveness ensuring no key aspects of the construct are missing, and comprehensibility to ensure items are understood as intended.[196] Feasibility is also key for integration into clinical practice.[196] Modifying pain assessment tools for children and young people with CP may improve content validity, feasibility, opportunity for self-report of the individual pain experience, and increase access to best practise pain interventions, thus improving quality of life.[209]

The primary objective of this mixed methods study, comprising of an online survey and a qualitative descriptive study, was to determine which of the currently available tools assessing impact of pain on emotional functioning would be most suitable to modify and/or validate in young people with CP. Secondary objectives were to identify

modifications required for improving appropriateness and feasibility of these tools, and to investigate content validity and feasibility of these tools in young people with CP.

4.5 Methods

Study design

This study was approved by the Women's and Children's Health Network Human Research Ethics Committee (2022/HRE00154) and was guided by COnsensus-based Standards for Measurement Instruments (COSMIN) recommendations for investigating content validity. Further, this study was informed by an advisory group of people with lived experience, including two young adults with CP and a parent of a child with CP.

Between January and April 2023, a two-stage mixed methods study was undertaken. In Stage 1, an online survey identified suitable tools for assessing the impact of pain on emotional functioning with potential for modification and/or validation. In stage 2, a qualitative descriptive study explored content validity, clinical feasibility and tool modification options. The consolidated criteria for reporting qualitative research (COREQ) were followed.[213]

Participants and recruitment

Participants in both stages were clinicians (physiotherapists, occupational therapists, psychologists, clinician researchers, speech pathologists, paediatricians, orthopaedic surgeons), individuals with CP (>8 years of age, the minimum age the tools had been developed for) and parents of children with CP (Table 4-1). All participants (stage 1 and 2) were recruited by online advertising and flyer distribution through the Australasian Academy for Cerebral Palsy and Developmental Medicine (AusACPDM), Novita, the Women's and Children's Hospital and the South Australian CP Register. Purposeful sampling was employed to facilitate inclusion of varied motor ability, communication ability and age in the qualitative component.[214]

Sample size

The expected maximum sample size for the survey was 60 clinicians and 30 people with lived experience, based on AusACPDM membership. For the qualitative study, the target sample size was 15-20 individuals, including at least 7 individuals with CP. COSMIN consider testing content validity in a qualitative study of seven or more participants from the population of interest as 'very good'.[30]

Table 4-1 Study eligibility criteria

Study component	Inclusion criteria	Exclusion criteria
Online survey	Individuals with a diagnosis of CP >8 years of age, any severity (GMFCS levels I-IV)	
	Parents or caregivers of children with CP (child age = 2-30 years)	
	Medical, nursing or allied health clinicians or clinician researchers with > 5 years' experience assessing and treating CP	Clinicians with < 5 years' experience assessing and treating CP
	English speaking and reading ability	
Qualitative descriptive study	Individuals with a diagnosis of CP >8 years of age, any severity (GMFCS levels I-IV)	
	Parents or caregivers of children with CP (child age = 2-30 years)	
	Medical, nursing or allied health clinicians or clinician researchers with > 5 years' experience assessing and treating CP	Clinicians with < 5 years' experience assessing and treating CP
	English speaking ability	Unable to communicate verbally or with a communication device in a focus group or interview setting

Procedure

Stage 1: Online survey

Survey data were collected and managed using REDCap electronic data capture tool, hosted at Murdoch Children's Research Institute.[215, 216] The survey was open for four weeks.

Survey instrument

The development of the survey included four main stages (Figure 4-1, Appendix C1 & C2). Available pain tools measuring impact of pain on emotional functioning were identified from three recent systematic reviews.[23, 24, 210] Pain coping tools were defined as those that assessed pain coping, including pain anxiety, pain catastrophising and fear of pain within a dedicated scale or subscale.[204, 205] Multidimensional tools were defined as assessing many pain domains in CP, with at least one item assessing impact of pain on emotional functioning, defined as "the extent to which pain hinders engagement with emotional functioning".[132] As advisory group members recommended no more than eight tools be presented in the survey, tools were reviewed for feasibility[27, 217] and the following tools identified for inclusion; three multidimensional tools (Modified Brief Pain Inventory (mBPI) [154], Pain Burden Inventory-Youth (PBI) [218], Pediatric Pain Screening Tool (PPST) [219]) and five pain coping tools (Bath Adolescent Pain Questionnaire (BAPQ) [220], Chronic Pain Acceptance Questionnaire (CPAQ) [221], Fear of Pain Questionnaire for Children-Short Form (FOPQ-C-SF) [222], Pain Vigilance and Awareness Questionnaire (PVAQ) [223] and Pain Catastrophizing Scale for Children (PCS-C) [224]).

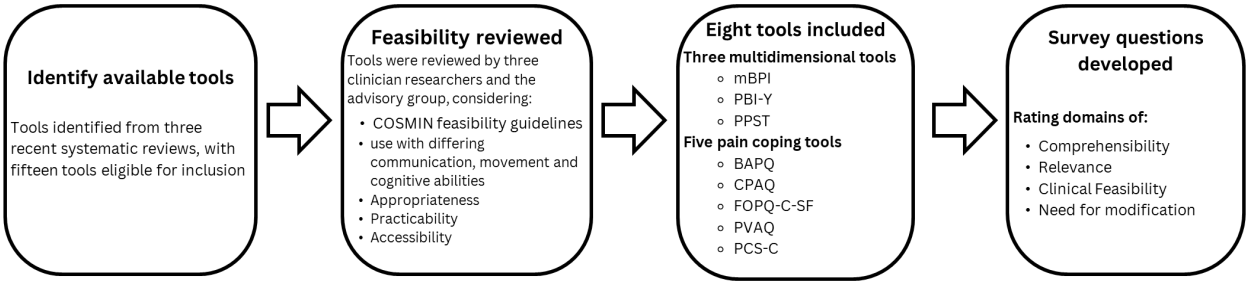


Figure 4-1 Development of the survey instrument
Further detail is provided in Appendix C1 & C2

***Abbreviations: bapq**=Bath Adolescent Pain Questionnaire, **cpaq**=Chronic Pain Acceptance Questionnaire, **fopq**=Fear of Pain Questionnaire for Children Short Form, **mbpi**=Modified Brief Pain Inventory, **pbi**=Pain Burden Inventory, **ppst**=Pediatric Pain Screening Tool, **pcs**=Pain Catastrophizing Scale for Children, **pvaq**=Pain Vigilance and Awareness Questionnaire*

Data analysis

Stage 1 determined the highest-rated tool for 1) pain coping and 2) multidimensional assessment, for further exploration in stage 2. Data were analysed by the percentage of 'strongly agree' and 'agree' responses for relevance, comprehensibility, clinical feasibility and need for modification of each tool (Appendix C3). Equal weighting was given across clinicians/researchers and lived experience participants, except for comprehensibility which was rated only by people with lived experience in line with COSMIN.[30]

Stage 2: Qualitative study

A qualitative descriptive study was undertaken to summarise the relevance, comprehensiveness, comprehensibility and feasibility of key tools assessing impact of pain on emotional functioning in children and young people with CP and chronic pain.[225, 226] Stakeholder recommendations to improve routine clinical use of chronic pain assessment tools were also identified.

Data collection methods

Adult participants were offered the choice of a focus group or individual interview, with separate groups for people with lived experience and clinicians. All children were interviewed to ensure they were able to express their views clearly.[227] Focus groups lasted 60 minutes and were conducted by a single interviewer (SK). Semi-structured interviews lasted 30-60 minutes and were conducted by a single interviewer (MGS). All focus groups and interviews were undertaken online using video conferencing software, digitally recorded and transcribed verbatim. Interviews with children (<18 years) were conducted with parents present, based on child/parent preference.

Participants were provided with a copy of the tools one week prior. Participants were advised they were able to cease or pause the interview at any time. A semi-structured interview guide was developed[227], and informed by both COSMIN requisites for content validity and stage 1 open-ended survey responses (Table 4-2, Appendix C4).[30, 31] The interview guide underwent rigorous testing prior to use: internal testing within the author team, and critique and field testing with the advisory group.[227]

Table 4-2 Key topics and prompts in the semi structured interview guide

Topic	Prompts	Participant group
Relevance	e.g. "Which questions are relevant to pain for people with cerebral palsy?"	All
Comprehensibility	e.g. "How could we make the questions easier to understand?"	All
Comprehensiveness	e.g. "Is there anything important or unique to people with cerebral palsy that is missing from this questionnaire?"	All
Clinical feasibility	e.g. "What suggestions would you make to improve the layout of this tool?"	All
Clinical feasibility	e.g. "How would you feel if you were asked to complete this tool before or during an appointment with your health professional?"	Parents/people with CP
Clinical feasibility	e.g. "How would you incorporate a tool like this into your clinical practice?"	Clinicians

Data analysis

Inductive content analysis was used to determine practical and concrete suggestions to inform practice.[226, 228] NViVO 12.0plus software (QSR International, Melbourne, Australia) was used to facilitate analysis. Briefly, an initial period of familiarisation was undertaken by two authors (MGS, ARH), whereby recordings were listened to, transcripts read and exploratory notes recorded. Coding was completed in two rounds, independently, by two authors (MGS, ARH). These authors met initially after completing two transcripts, then regularly to compare categories applied and to ensure consistent terminology. In the first round, transcripts were coded to identify big picture meaning units and develop a preliminary big picture coding schema.[228] The second round coding used a 'line by line' process to produce subcategories, resulting in a refined coding schema (Appendix C5), which was reviewed by an external qualitative advisor (AC). Each suggested modification was identified and mapped to the big picture categories.

Researcher characteristics and reflexivity

Inductive content analysis recognises the researcher's perspectives and prior experiences may influence data analysis.[228] MGS is a female PhD student and physiotherapist, and has training in qualitative research. SK is an experienced female qualitative researcher and education specialist. ARH, RR and RJG supervised the research. ARH is an experienced disability researcher, physiotherapist and also has training in qualitative research. RR is a paediatric rehabilitation medical specialist and researcher, with >30 years' experience working with children with CP. RJG is an experienced researcher with >20 years' experience in both quantitative and clinical studies.

Trustworthiness

Credibility was demonstrated by establishing rapport with patients prior to commencing interviews, with some participants already known to MGS through clinical relationships. Transcripts and quotes were reviewed by participants. Using focus groups alongside interviews reduced the role of the researcher, improving authenticity. Including participant demographics to demonstrate the breadth of age, communication ability, cognitive ability and motor ability represented in the study, establishes transferability. Direct quotes were included to ensure confirmability of the findings. Qualitative data, including the final coding schema, were triangulated with online survey open-ended responses and advisory group.

4.6 Results

Participant characteristics

Sixty-one participants completed the online survey (32 clinicians, 19 parents and 10 individuals with CP) and 30 participants undertook the qualitative descriptive study (12 clinicians, 9 parents and 9 young people with CP [12-39 years of age]) (Table 4-3). No participants withdrew from the study.

Table 4-3 Descriptive statistics of participants

	Online survey (n=61)	**Qualitative descriptive study (n=30)**
	% (n)	% (n)
Clinicians	**52.5% (32)**	**40% (12)**
Occupational Therapist	*1.6% (1)*	-
Orthopaedic Surgeon	*3.2% (2)*	-
Paediatrician	*6.6% (4)*	*3.3% (1)*
Physiotherapist	*36.1% (22)*	*26.7% (8)*
Psychologist	*1.6% (1)*	*3.3% (1)*
Speech Pathologist	*1.6% (1)*	*3.3% (1)*
Other (clinician researcher)	*1.6% (1)*	*3.3% (1)*
People with lived experience of CP	**47.5% (29)**	**60% (18)**
Young people with CP *[Age range]*	*16.4% (10)^*	*30% (9)* *[12-39 years]*
Parents of children with CP *[age range of child]*	*31.1% (19)*	*30% (9)* *[4-25 years]*
Mobility Level (percentages based on the lived experience participants only)		
	n=29	**n=18**
GMFCS I	*13.8% (4)*	*5.6% (1)*
GMFCS II	*41.4% (12)*	*50% (9)*
GMFCS III	*17.2% (5)*	*11.1% (2)*
GMFCS IV	*6.9% (2)*	*11.1% (2)*
GMFCS V	*20.7% (6)*	*22.2% (4)*
Communication ability (percentages based on lived experience participants only)		
	n=29	**n=18**
Able to report and describe pain without any additional assistance	*75.9% (22)*	*72.2% (13)*
Able to report and describe pain with the use of a communication device or other method	*6.9% (2)*	*11.1% (2)*
Unable to report and/or describe pain	*17.2% (5)*	*16.7% (3)*
School type (percentages based on the child participants only n=3)		
Mainstream		*66.7% (2)*
Special unit		*33.3% (1)*

^No children with CP (<18 years) participated in the online survey, age data for >18 years was not collected for the survey

Stage 1: Online survey

The FOPQ-C-SF was the highest rated pain coping tool for both relevance and clinical feasibility (Figure 4-2, Appendix C6) whereas the mBPI was the highest rated multidimensional tool for both relevance and clinical feasibility (Figure 4-3). Both the FOPQ-C-SF and mBPI were second highest rated for comprehensibility (Figure 4-2, Figure 4-3).

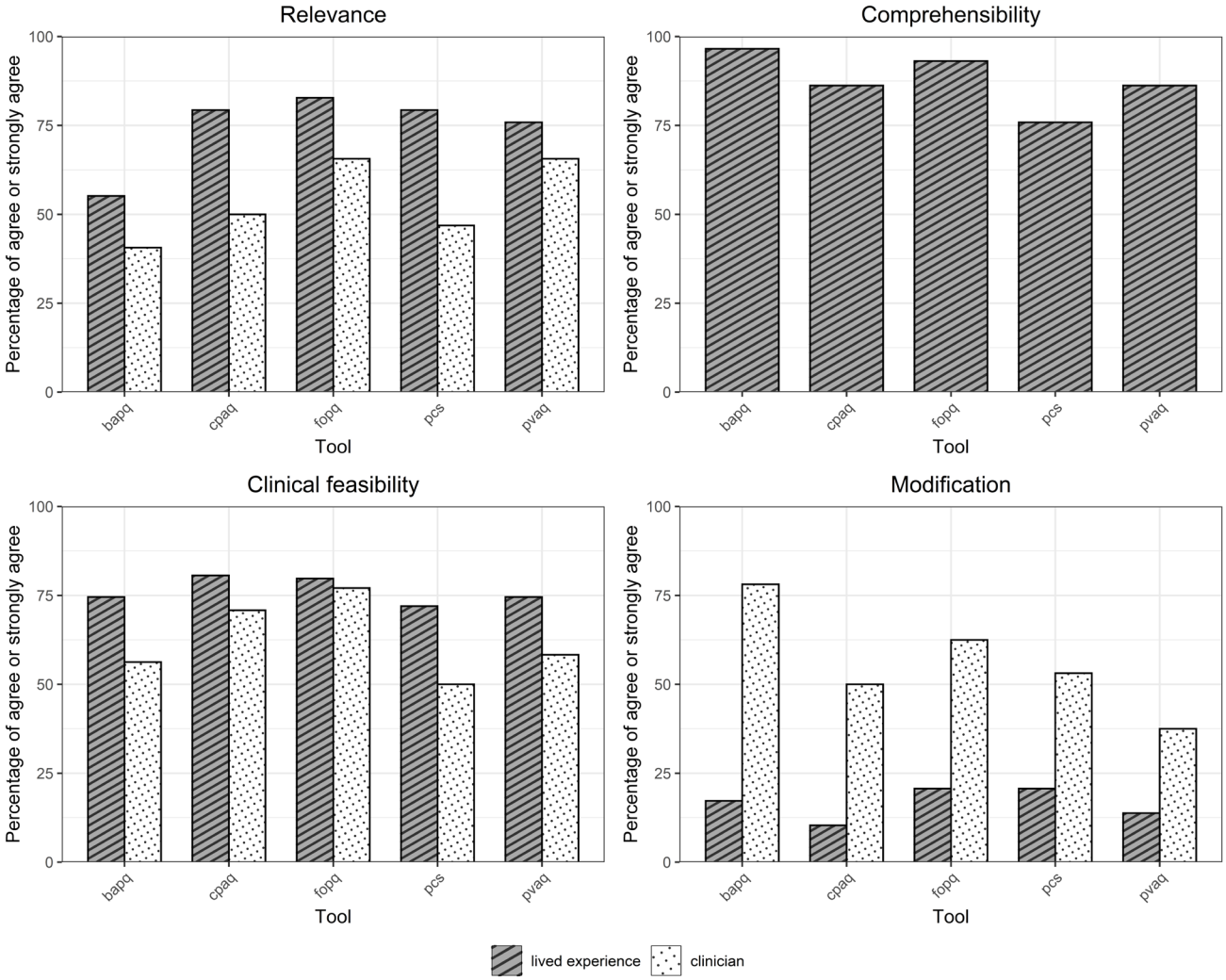


Figure 4-2 Ratings of relevance, comprehensibility, clinical feasibility and the need for modification of pain coping tools

Abbreviations:* bapq=Bath Adolescent Pain Questionnaire,* **cpaq=***Chronic Pain Acceptance Questionnaire,* **fopq=***Fear of Pain Questionnaire for Children Short Form,* **pcs=***Pain Catastrophizing Scale for Children,* **pvaq=***Pain Vigilance and Awareness Questionnaire*

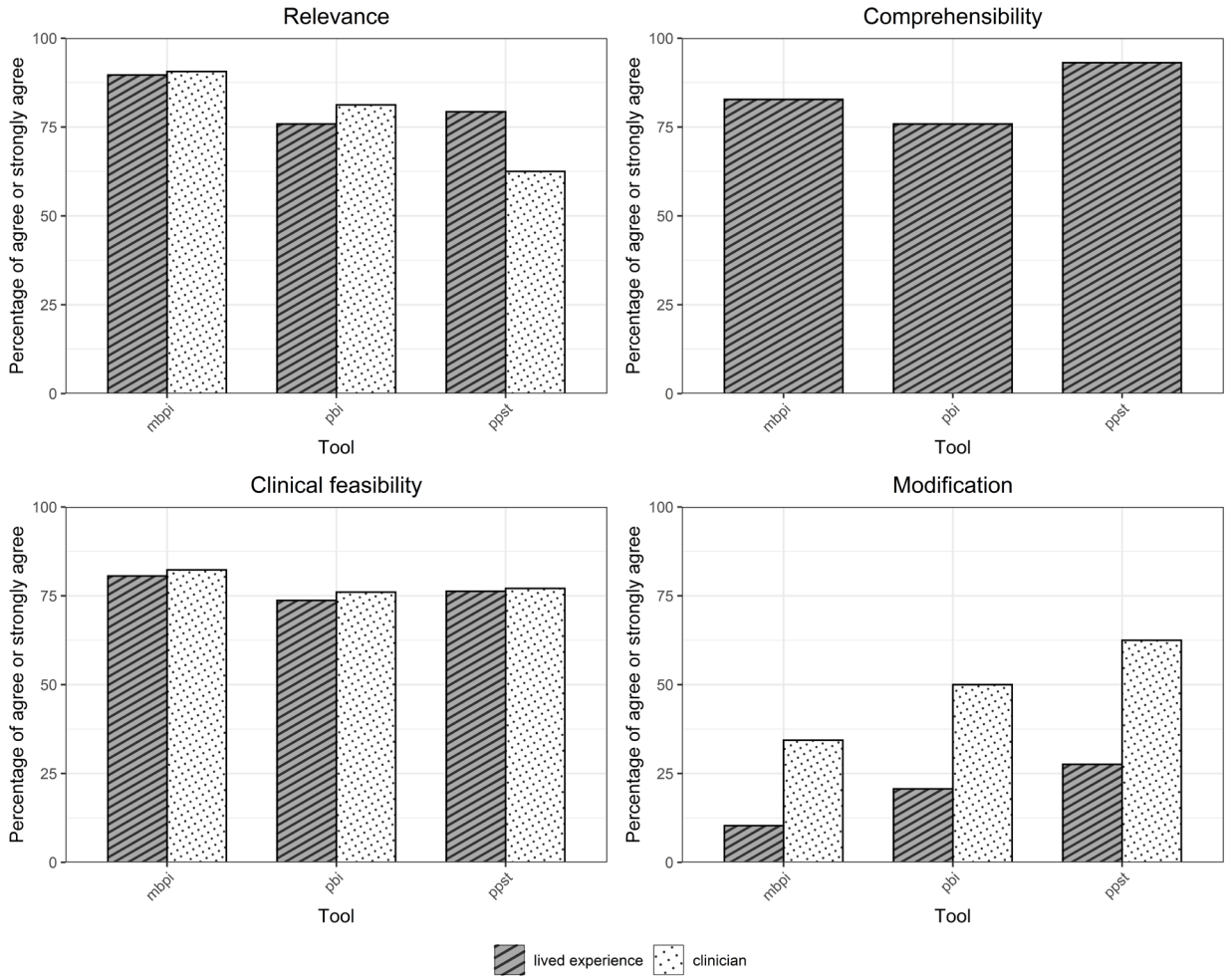


Figure 4-3 Ratings of relevance, comprehensibility, clinical feasibility and the need for modification of the multidimensional tools for chronic pain including assessment of 'impact of pain on emotional functioning'

***Abbreviations: mbpi**=Modified Brief Pain Inventory, **pbi=**Pain Burden Inventory, **ppst=**Pediatric Pain Screening Tool*

Stage 2: Qualitative study

Five focus groups (three lived experience and two clinician groups) and eight interviews (two clinicians, one parent and five people with CP) were completed. Participant data were coded to six categories: accessibility, comprehensibility, feasibility, relevance, presentation and comprehensiveness (Figure 4-4, Table 4-4). People with lived experience provided feedback primarily focused on relevance and comprehensibility, while clinicians provided feedback focused on presentation and comprehensibility. Fifty-eight unique modification suggestions were identified, 14 by people with lived experience only and 17 by clinicians only (Appendix C7). The remaining modification

suggestions were identified by both groups. A full list of quotes is provided in Appendix C8. Representative quotes are formatted as follows:

Experience type: age/child age: GMFCS level: communication ability (able to self report or complex communication needs (CCN))

Table 4-4 Qualitative data category and subcategory description

Category	Category description	Subcategories	Further subcategories
Accessibility	Suggestions for how people with disability can access the tool to prioritise self-report	• Cognitive impairment • Complex communication needs	
Comprehensibility	Comments related to an individual's ability to understand the tool - including but not limited to understanding the items, wording, purpose of the tool and response options	• Age • Co-occurring diagnosis • Unclear wording • Understanding what pain is	
Comprehensiveness	Suggestions to ensure the full scope of the construct is covered, as is relevant to people with cerebral palsy. The focus here is on clarifying existing items, not adding new items		
Feasibility	Suggestions related to the use of the tool in clinical practice and research by clinicians and people with lived experience	• Administration • Appropriateness of parent report • Emotional response • Purpose of the tool	*Validity of the assessment*
Presentation	How the assessment tool is displayed or presented	• Scaling • Visual presentation	
Relevance	Suggestions relating to the relevance or meaning of the tool for people with cerebral palsy	• Age group • Inappropriate wording • Meaningful to children with CP • Purpose of items • Purpose of tool • Recall period	*Autonomy*
Suggested changes	All suggested practical changes/modifications to the tools		

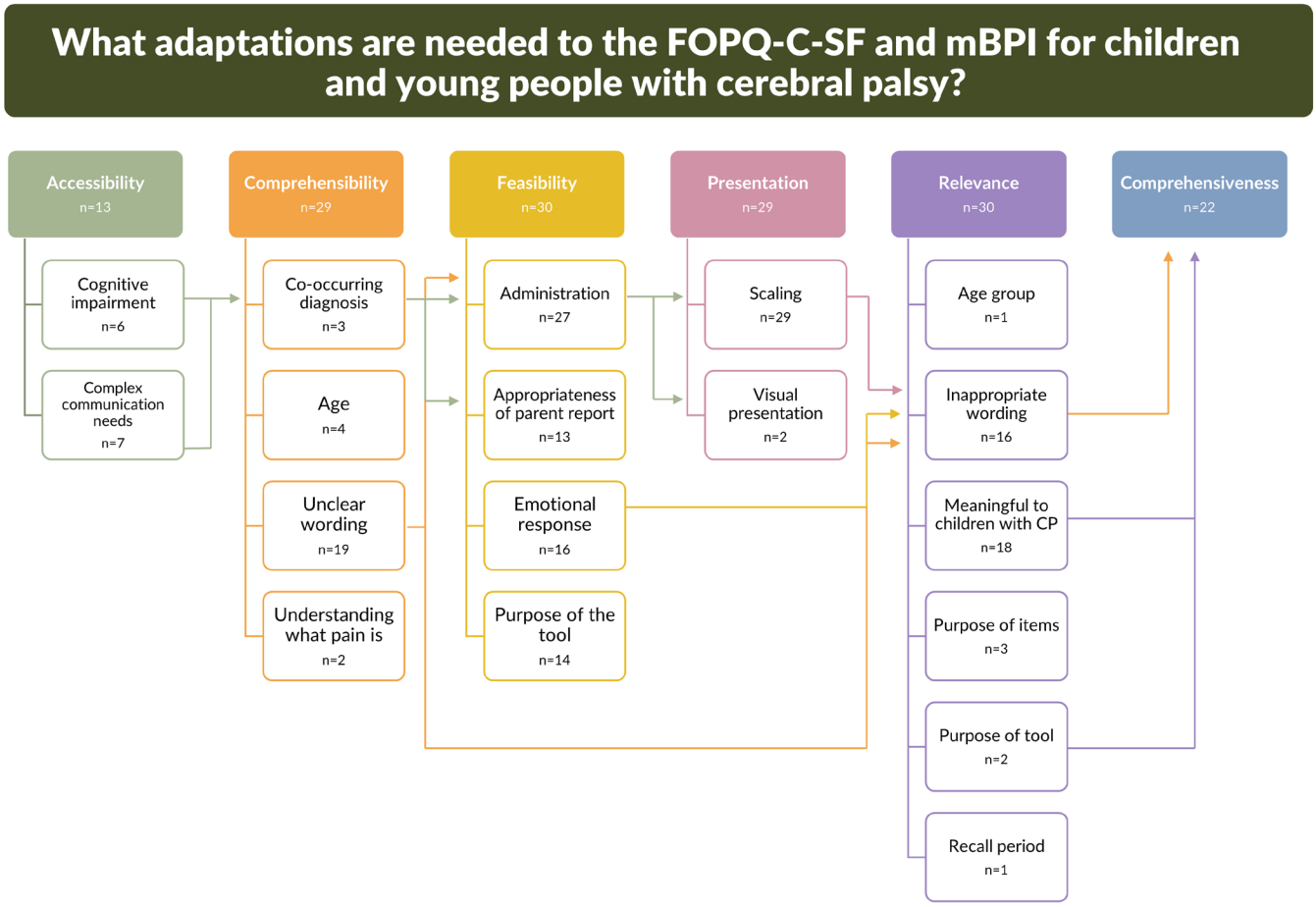


Figure 4-4 Categories from the qualitative descriptive study.

Number of participants contributing data to each category is depicted by n= X

Comprehensibility

Comprehensibility, related to the ability to understand the tool itself, individual items, wording and response options. Suggestions included simplifying mBPI wording, with the FOPQ-C-SF identified as already using appropriate language. Improving comprehensibility involved using alternatives to written text, particularly for those with cognitive impairment and of younger age.

> *"I think... with younger kids, especially non-verbal, [you should]* ***use a lot of visuals*** *because they understand through that a lot easier..... you know, or even someone who is verbal, like [my daughter], sometimes,* ***visuals are a lot better for her to understand*** *rather than asking her questions and getting her to answer."*
>
> *(Parent, 9-year-old female, GMFCS II, able to self-report)*

Parents and children with CP acknowledged they did not always understand what pain was because it was difficult to identify and pain was a normal part of life. Clinicians highlighted the potential difficulty for children and young people with co-occurring diagnoses of CP and Autism using a tool with abstract questions, such as the FOPQ-C-SF.

> *"Because when I look at that question, when I feel pain I'm afraid that something terrible will happen, I just wonder how that will be interpreted.... Because it's also got to take into account their [individuals with Autism] ability to introspect and to understand feelings as well as pain, which they may have difficulties doing"*
>
> *(Psychologist)*

Accessibility

Accessibility observations primarily related to access for self-report for people with cognitive impairment and complex communication needs. Suggested improvements included using alternatives to written text, different ways to administer the tool including asking questions one at a time, having one question per page, using a *Talking Mats®* framework[229] and providing an option for 'I don't know':

> *"So **if you are verbal**, you can say, I don't understand what you asked me or what do you mean or you can ask questions back and forth to clarify but **if you are just expected to point to one to 10 or whatever you make the scale,** you need some of that interactive language for them to be able to say, like**, "explain this to me more", "I don't understand", "I don't know**." Because **how do you opt out of a scale like that?"***
>
> *(Speech Pathologist)*

Participants also recognised making accessibility changes to the tools needed to be individualised:

> *"Yeah, I think **simpler wording**. If we could **base it around his PODD** [it] would probably make it much easier. But then it's kind of very*

customised and that's going to take a lot of work to ***specifically customise it to him****."*

(Parent, 8-year-old, GMFCS V, CCN)

Comprehensiveness

Comprehensiveness focused on ensuring the full scope of the construct was covered as is relevant to people with CP. Suggestions included providing examples of the item with standardised examples or descriptions:

"I think you would either list them all as an explainer as to what it all means or you have a separate one for each..... Daily care needs, I think, even if it's for a mother... still needs to be specific. You need to put in brackets, washing, brushing teeth, getting dressed. I still think you need a little bit of clarification on what that means"

(Parent, GMFCS V, CCN)

Participants suggested including high frequency events specific to children and young people with CP, such as asking how pain interferes with use of assistive technology and pain related fear in the context of therapy, equipment, medical interventions or visiting health professionals.

"When she [my daughter] sees her physiotherapist, you can just see the fear in her eyes straightaway because you know the physio is going to put her through her paces a bit and make her uncomfortable through the various stretches and exercises and all that type of thing."

(Parent, 9-year-old, GMFCS V, CCN)

Presentation

Presentation most commonly related to scaling/response options and visual presentation, closely related to accessibility and comprehensibility. All participants agreed on reducing the number of response options in the mBPI. Reducing the scale to 5 points, with visual symbols, verbal descriptions and colour scales for each of the numbers, was suggested.

"Maybe one to five instead of one to 10.....it's a large scale to be choosing from... there's not probably a lot of difference between four and five, five and six"

(39-year-old, GMFCS II)

The most commonly suggested visual symbols for the mBPI were faces showing emotions, or symbols showing empty to full. For the FOPQ-C-SF, the most commonly suggested symbol was thumbs up/thumbs down to indicate level of agreement. The importance of having a range of options for different people was highlighted, particularly having visual symbols, words and colours available together or separately on different versions. Participants also recommended larger font, bigger spacing and alternating colour rows to improve readability.

"I think if you're going to get the person with CP to fill it out though, you might want to lay it out slightly differently. Because obviously, if you've got trouble with fine motor skills, circling a small number is going to be quite difficult. So even if you had it in like boxes that were larger and had the wording like does not interfere and completely interferes at either end rather than one underneath and one to the side and maybe some more spacing between them would be a lot easier"

(Parent, 4-year-old, GMFCS III, able to self-report)

Feasibility

The feasibility of tools was identified by all participants. Suggestions related to when and how the tool is completed, appropriateness of parent report, possible emotional responses by those completing it and understanding the tool purpose. Presenting items one at a time, ensuring understanding and then selecting a response option was the preferred method. People with lived experience wanted to complete the assessments before seeing a clinician, with children preferring to complete the assessment with a parent/caregiver.

People with CP and parents both indicated a proxy report version of the FOPQ-C-SF was not appropriate because pain-related fear is too personal to be answered accurately by

a proxy, even someone who knows the individual well. Pain-related fear was the only construct discussed that was identified as being too personal for a proxy report.

People with lived experience acknowledged their frustration at completing assessments without feedback from the clinician afterwards.

> *"Because from my previous experience [they] always ask me to fill the questionnaire, **but no update afterwards**. So that's why sometimes I just feel like, you know, they say, oh, this is very important to us, it would be great if you filled this out, blah blah blah. We do it, we take our time and we do it seriously, but sometimes **we just feel like we didn't hear any feedback since then**. So, it would be great that everything you do for [a] purpose. **So, it would be great if I just know what's happening**, even though maybe nothing really happened, and then you can let us know that you do it and you read it"*
>
> *(Parent, 10-year-old, GMFCS II, CCN)*

All groups recognised answering questions about pain could be a negative experience, highlighting the importance of clinicians knowing when a tool like this was necessary, and explaining this to the client.

Relevance

Relevance was identified by all participants. Alternative wording for people with disability was frequently suggested, particularly in the context of movement (i.e. using "mobility" not "walking/running"). The phrase "normal people" (item 1 FOPQ-C-SF) was seen as inappropriate for individuals with disability.

> *"The tricky one I see straight away is, can I do all of the [things] normal people do.... because it's easy to hurt my body? It's not about what people can do, because... you struggle with motor skills. Some of those questions are strange, because it'll be like, you can't kick a ball, because your back hurts? Well, yeah, but I can't actually kick the ball.... probably just ditch normal people. Or you can just put people. I personally don't really care – I don't know if I've been desensitised, or*

whatever, but I know people can get a little bit jarred with that [normal people].”

(16-year-old, GMFCS II)

All groups acknowledged the FOPQ-C-SF had an assumption of autonomy and independence of the respondent, which may not be true for children with CP. Wording changes were suggested to reflect this.

“[Children without disability] can move – like, if you are mobile, if someone comes at you with an injection, you bolt across the room, and you hide behind your mum, and you would be kicking and screaming. Whereas, if you can't do that, yeah, you might be crying, but people ignore that. So, yeah, then you…start to develop….fear about all those things, because you're not in control at all”

(Speech pathologist)

4.7 Discussion

This study highlighted the importance of involving clinicians and people with lived experience in the modification of assessment tools for young people with CP and chronic pain. The FOPQ-C-SF and mBPI were the highest rated tools assessing pain coping and multidimensional assessment of the impact of pain on emotional functioning (respectively). Potential tool modifications were identified, relating to six categories: accessibility, comprehensibility, feasibility, relevance, presentation and comprehensiveness.

COSMIN recommends, where possible, existing assessment tools are tested and used in new populations rather than developing new tools.[31] Accordingly, we focused on validating and modifying existing tools for young people with CP. Suggestions which changed the construct of interest were not considered.[31] The eight tools included in the survey covered four main constructs: pain catastrophising, pain anxiety, pain-related fear, and pain interference with emotional functioning.[132, 204, 205] Despite high ratings of relevance, comprehensibility and feasibility across the eight tools, we chose to start by modifying the two highest-rated, which assess two constructs: fear of pain (FOPQ-C-SF) and pain interference, including pain interference with emotional functioning (mBPI).

These modifications could also be applied to other future outcome measures, enabling assessment of a wider range of pain-related constructs. This study also included lived experience representation of cognitive and communication impairment to ensure accessibility of modifications.

Heterogeneity is a challenge when ensuring tools can be used in the CP population. Young people with CP can have varied motor, communication, visual, sensory and/or cognitive abilities.[57] Modifying tools to cater to all needs in the population is difficult. For example, some participants suggested adding a colour scale to improve understanding for those with cognitive impairment, whereas others disagreed because for those with visual impairment, added colours can increase confusion.

A further challenge is balancing the needs of clinicians and people with lived experience. While both groups agreed on which tools to use, differing views were held on how tools should be presented. For example, people with lived experience requested each question be displayed on a separate page, however clinicians perceived this to create additional workload by needing to collate multiple pages. Although this may be an impracticality for clinicians, modifications to improve self-report opportunity for people with lived experience should be prioritised given they are the ones completing the tools and 1 in 2 children with CP have an intellectual disability.[57] The need for adaptation in the survey was lower for the lived experience group than clinicians, despite similar numbers of changes suggested by the groups in the qualitative component. The advisory group suggested this may be because people with lived experience are used to 'making do' with inappropriate assessments, not because they do not need modification.

Consumer engagement is fundamental to ensure knowledge translation.[230] The advisory group was instrumental in recommending procedures to ensure representation of the heterogenous population, while reducing participant research burden. Both the quantitative and qualitative components had close to equal representation of clinicians and people with lived experience, with lived experience participants being representative of the spectrum of abilities seen in CP.

A limitation of this study was possible bias as the interviewer MGS was known to some participants. A further limitation was not interviewing children with moderate-severe cognitive impairment or complex communication needs. To gather modification suggestions, participants needed to understand the existing assessment tools and suggest changes. We instead focused on ensuring representation of these groups through proxy reporters, such as parents.

Future directions

Identified modifications to the FOPQ-C-SF and mBPI will be further investigated to determine which are most important to implement. Further testing of comprehensibility, through pilot testing and cognitive interviews, will then take place in young people with CP and varying movement, communication and cognitive abilities. The identified modification suggestions could also be used to adapt other outcome measures for young people with CP.

4.8 Conclusion

The FOPQ-C-SF and mBPI are the highest rated tools for assessing impact of pain on emotional functioning for children and young people with CP, provided modifications are made. Potential modifications have been identified to improve appropriateness and feasibility for children with CP and varying abilities. Future research should prioritise the involvement of people with lived experience when modifying PROMs to ensure their needs are balanced alongside those of clinicians.

Chapter 5 – Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study (Study 3)

5.1 Preamble

Chapter 4 reported the results of my mixed methods study which identified potential modifications to the two highest rated tools for assessing the impact of pain on emotional functioning for children and young people with CP: the modified Brief Pain Inventory (mBPI) (multidimensional tool) and the Fear of Pain Questionnaire for Children-short form (FOPQ-C-SF) (dedicated scale). It should be noted that the Patient-Reported Outcomes Measurement Information System (PROMIS) Pediatric Pain Interference Scale was not presented as part of the selection and modification suggestion process. PROMIS is a world-wide approach aiming to improve consistency and quality of patient-reported outcome measurement, however the strict development and testing process, which uses Item Response Theory (IRT) methods, requires large participant numbers (>500) and was therefore outside of the scope of this thesis.

Fifty-eight unique modification suggestions to the mBPI and FOPQ-C-SF were identified within six broad categories of comprehensiveness, comprehensibility, relevance, accessibility, presentation and feasibility.[211] However, several of the suggestions were conflicting and the breadth of the suggestions made immediate implementation difficult. For example, one parent identified using a colour scale would improve comprehensibility for their child with CP, however another parent advised a colour scale would make it less accessible for their child due to visual impairment. Although this was not an unexpected finding given the heterogeneity of the CP population[57], it did highlight the need to reach agreement on which modifications were most important to implement.

The COSMIN content validity guidelines do not provide specific recommendations for this circumstance.[30] The project advisory group strongly suggested the involvement of a wider group of clinicians and people with lived experience to determine which modifications should be implemented, using a consensus process.[231] This was particularly important given the goal of enabling broad use of the adapted tools in

research and clinical practice. Accordingly, I designed a consensus process to include a range of people with lived experience, including representation of young people with diverse cognitive, communication and functional abilities, along with clinicians working with young people with CP.[232]

Chapter 5 reports the results of my two-round modified electronic Delphi study, with the primary aim of reaching consensus on which of the modification suggestions to the two tools (mBPI and FOPQ-C-SF) should be implemented. This manuscript has been published in Pain Reports.

5.2 Authorship declaration

This manuscript has been published in Pain Reports

Smith, M.G.[1], Gibson, R.J.[1], Russo, R.N.[2,3], Harvey, A.R[4,5]. Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study. *Pain Reports* 10(4):p e1304, 2025. https://doi.org/10.1097/pr9.0000000000001304

[1]School of Allied Health Science and Practice, University of Adelaide, Adelaide, Australia
[2]College of Medicine and Public Health, Flinders University, Adelaide, Australia
[3]Paediatric Rehabilitation Department, Women's and Children's Hospital, Adelaide, Australia
[4]Neurodisability and Rehabilitation, Murdoch Children's Research Institute, Melbourne, Australia
[5]Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia

Statement of Authorship

Title of Paper	Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study
Publication Status	X Published ☐ Accepted for Publication ☐ Submitted for Publication ☐ Unpublished and Unsubmitted work written in manuscript style
Publication Details	Smith, M.G., Gibson, R.J., Russo, R.N., Harvey, A.R. Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study. *Pain Reports* 2025. 10:e1304. https://doi.org/10.1097/pr9.0000000000001304

Principal Author

Name of Principal Author (Candidate)	Meredith Smith		
Contribution to the Paper	Completed data collection, data analysis, wrote manuscript and acted as corresponding author		
Overall percentage (%)	70%		
Certification:	This paper reports on original research I conducted during the period of my Higher Degree by Research candidature and is not subject to any obligations or contractual agreements with a third party that would constrain its inclusion in this thesis. I am the primary author of this paper.		
Signature		Date	6/1/25

Co-Author Contributions

By signing the Statement of Authorship, each author certifies that:

i. the candidate's stated contribution to the publication is accurate (as detailed above);

ii. permission is granted for the candidate in include the publication in the thesis; and

iii. the sum of all co-author contributions is equal to 100% less the candidate's stated contribution.

Name of Co-Author	Adrienne Harvey		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	13-01-2025
Name of Co-Author	Rachel Gibson		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	10/2/25
Name of Co-Author	Ray Russo		
Contribution to the Paper	10% Supervised development of work, edited manuscript		
Signature		Date	6/1/2025

5.3 Abstract

Introduction: Cerebral palsy (CP) is the most common cause of childhood physical disability. Chronic pain is a common comorbidity of CP, however holistic pain assessment is challenging due to few available self-report tools for people with CP and diverse cognitive, communication and functional abilities. Previous research has identified two tools for potential use in CP (Fear of Pain Questionnaire for Children-Short Form (FOPQ-C-SF) and Modified Brief Pain Inventory (mBPI)), along with potential modifications.

Objectives: To gain consensus on modifications required to the FOPQ-C-SF and mBPI for use with children and young people with CP and chronic pain.

Methods: A two-round modified electronic Delphi study with people with lived experience (n=19 and 16) and clinicians (n=25 and 21) was conducted. Ethics was obtained through the Women's and Children's Health Network Human Research Ethics Committee (2022/HRE00154). A total of thirty-seven modification suggestions to the FOPQ-C-SF and mBPI were rated on a 5-point Likert scale (strongly agree to strongly disagree). Consensus was determined as ≥75% of participants rating the suggestion as strongly agree/agree, or strongly disagree/disagree.

Results: Seven modifications reached consensus for inclusion in the FOPQ-C-SF and 21 for the mBPI. No modifications reached consensus for exclusion for either tool. Modifications primarily focused on refining wording; enhancing visual representation of the tool for improved understanding and reducing the number of response options**.**

Conclusion: Consensus has been reached on modifications to the FOPQ-C-SF and mBPI for young people with cerebral palsy. Future research will examine the comprehensibility of the adapted tools across the population.

5.4 Introduction

Cerebral palsy (CP) is the leading cause of childhood physical disability worldwide, with a prevalence of approximately 1.6/1000 live births.[1] Chronic pain, defined as "persistent or recurring pain lasting longer than three months", is more frequently experienced by children and young people with CP than other paediatric populations.[3] It is well established that chronic pain should be assessed within a biopsychosocial framework, acknowledging the broad impact of chronic pain on a person's life.[20] Twelve core pain assessment domains have previously been identified for children and young people with CP.[233] However, few tools which assess pain have evidence of validity for use in CP, or are appropriate for the whole spectrum of abilities seen in CP; for example communication, cognitive, visual and motor abilities.[24, 210] Without access to valid and appropriate pain assessment tools, children and young people with CP have limited opportunity to access best practice interventions for chronic pain.[34]

Previous research has identified a need for tools assessing the impact of pain on emotional functioning for children and young people with CP.[23, 24, 210] The Pediatrics Initiative on Methods, Measurement and Pain Assessment in Clinical Trials (PedIMMPACT) paediatric chronic pain domains specify emotional functioning, such as anxiety and depression, as a core area of assessment for children with chronic pain.[234] When core assessment domains were developed specifically for children and young people with CP, this was refined to 'impact of pain on emotional functioning' to better distinguish pain-related emotional functioning and general emotional functioning.[203] Factors contributing to the impact of pain on emotional functioning, such as pain coping, influence pain-related physical disability, anxiety and depression [204, 205] and are also key targets for pain-related psychological interventions.[206] Previous work by our team sought to identify which tools focusing on the impact of pain on emotional functioning may be suitable for children and young people with CP.[211] Briefly, an online survey with clinicians and people with lived experience of CP was conducted to rate the relevance, comprehensiveness, feasibility and comprehensibility of tools in two groups: firstly, tools that assessed the impact of pain on emotional functioning in a dedicated scale or subscale and secondly, multidimensional tools with at least one item assessing the impact of pain on emotional functioning. The two highest rated tools were the Fear of Pain Questionnaire for Children-Short Form (FOPQ-C-SF) (dedicated scale);

and the modified Brief Pain Inventory (mBPI) (multidimensional).[211] The FOPQ-C-SF was developed by Heathcote et al. (2020)[235] as a shortened version of the Fear of Pain Questionnaire for Children[236], assessing pain-related fear and providing an understanding of the possible impact on pain-related disability.[236] The Brief Pain Inventory (BPI) was originally developed for adults with cancer pain to measure pain interference, pain intensity, changeable factors and pain location.[237] In 2009, Engel et al.[238] modified the pain interference items of the BPI to be more suitable for children with neuromuscular disorders. The modified version of the BPI was subsequently tested in parents of children with CP as a proxy report measure.[154] Although both assessment tools have been used in children with CP, until recently there was no investigation of content validity in this population.[24, 210]

Our recent study sought to address this gap by investigating the content validity and feasibility of both tools for children and young people with CP and chronic pain.[211] Briefly, in order to improve relevance, comprehensibility and accessibility of the tools across the spectrum of ability, we conducted a qualitative descriptive study with people with lived experience and clinicians. We identified the FOPQ-C-SF and mBPI could be useful in their current form for youth with CP, but only those without cognitive or communication impairment.[211] The study showed the tools required adaptation of the content to ensure they were relevant and comprehensible for all children and young people with CP. For example, the wording of the mBPI was considered too complex for children or those with cognitive impairment, with phrases like ‘recreational activities’. Furthermore, the FOPQ-C-SF wording did not reflect the potential reduction in autonomy a person with a disability might experience when they are unable to stop an activity due to external limitations, such as needing assistance to get out of a piece of equipment. Fifty eight unique modification suggestions were identified from this study; however a number of the suggestions were conflicting, and the breadth of the recommendation suggestions made implementation difficult. For example, one parent identified using a colour scale would improve comprehensibility for their child with CP, however another parent advised a colour scale would make it less accessible for their child due to visual impairment. Although this was not an unexpected finding given the heterogeneity of the CP population[57], further research was recommended to gain consensus on which modifications should be implemented.[211]

Therefore, the primary objective of this current study was to gain consensus among people with lived experience and clinicians on which modifications to the FOPQ-C-SF and mBPI should be implemented to ensure appropriateness for children and young people with CP and chronic pain.

5.5 Methods

Study design

We selected a two-stage modified electronic Delphi study, conducted between July and September 2023, to facilitate expert consensus on modifications to the tools that had been proposed and discussed in a previous focus group setting, but had not yet reached a conclusion.[203] A standard Delphi begins with an open-ended question round to generate items, whereas a modified Delphi, as used here, starts with a structured questionnaire based on existing literature.[239] In this study, the initial survey round was informed by findings from the preceding qualitative study.[211] The asynchronous nature of the electronic Delphi design also provided greater flexibility, allowing for broader participation from both people with lived experience and clinicians. The study design was guided by COnsensus-based Standards for Measurement Instruments (COSMIN) recommendations.[240] Our study was co-designed and conducted with a consumer advisory group; including two young adults with CP and a parent of an adolescent with CP. The consumer advisory group was involved in defining the study purpose, piloting data collection tools and reviewing data analysis methods.[226]

Ethics approval was granted through the Women's and Children's Health Network Ethics Committee (2022/HRE00154). All participants provided informed consent, obtained through a consent statement in the survey.

Participants and recruitment

Participants were recruited nationally and internationally to form two groups: group 1 - clinicians (orthopaedic surgeons, paediatricians, physiotherapists, psychologists, speech pathologists, clinician researchers) and group 2 - people with lived experience (children with CP, adults with CP and parents of children with CP). Participants were eligible for inclusion if they were individuals with a diagnosis of CP (>8 years of age); parents or caregivers of children with CP (child age 2-30 years) and medical, nursing or

allied health clinicians or clinician researchers with >5 years' experience working with children and young adults with CP. Participants were recruited through online advertising and email distribution throughout known networks, including the National Research Group for Pain in Cerebral Palsy, the Australasian Academy for Cerebral Palsy and Developmental Medicine, Novita, the Women's and Children's Hospital Paediatric Rehabilitation Department and the South Australian Cerebral Palsy Register. All participants were screened with an online demographic survey to ensure they met the inclusion criteria.

Sample size

Although there are no specific guidelines when selecting participant size for a Delphi study, research suggests a minimum of 12 participants is required to reach consensus.[241, 242] For this study, it was important to ensure clinicians had sufficient expertise working with clients with CP (defined as >5 years' experience), as well as ensuring people with lived experience were represented from across the spectrum of mobility and communication ability within CP. To ensure people with lived experience had strong representation in the study, we determined that at least one third of the study participants should be people with lived experience. We determined a maximum of 60 clinicians/researchers and 30 people with lived experience might be recruited for this study, utilising the methods described. These numbers were based on the membership of the Australasian Academy for Cerebral Palsy and Developmental Medicine (AusACPDM), with fourteen members in the national research group for Pain in Cerebral Palsy.

The survey instrument

Survey questions were developed based on the modification suggestions identified by our previous study.[211] Modification suggestions were reviewed by all authors and the consumer advisory group, and grouped into a) modifications to be presented in the Delphi study for consensus, b) modifications which could be implemented without requiring consensus; for example changes to the administration mode of the tool rather than changes to the tool itself, this included adding alternative methods of access (e.g.

audio) and c) modifications which were considered inappropriate to take further. Detailed information on the modifications in each group is provided in Appendix D1. Modifications that were not taken further included those that would change the construct of interest of the tool. A final list of 38 suggestions were included in the survey instrument (Appendix D2). During data analysis, one further suggestion was identified as changing the construct of interest and was removed from the analysis (Wording change – FOPQ-C-SF- item 9: "I stop any activity if I start to hurt or my pain becomes worse" to "pain stops me doing things I want to do"). A total of 37 suggestions were included in the analysis (eleven relating to the FOPQ-C-SF and 26 relating to the mBPI).

Survey protocol

All potential participants were emailed information regarding the project with a link directly to the electronic Delphi survey. Survey data were collected and managed using REDCap electronic data capture tool, hosted at Murdoch Children's Research Institute.[215, 216] Each round was open for four weeks. In round 1, participants were presented with the original FOPQ-C-SF and mBPI, and asked to rate each of the suggested changes on a five-point Likert scale (strongly agree – strongly disagree). An optional 'comments' box was included, allowing participants to provide unprompted, open-ended feedback on each individual suggested change.. Participants who completed round 1 were subsequently emailed round 2. This round identified modifications which did not reach consensus and participants were asked to re-rate these. Participants were given data on which modifications reached consensus in round 1, along with graphs displaying the percentage responses (strongly agree, agree, neutral, disagree and strongly disagree) for each modification that did not achieve consensus. The graphs clearly separated the responses from clinicians and people with lived experience, allowing participants to see the differences between the two groups. Up to three reminder emails were sent for round 2. Modifications which reached consensus for one group but not the other (i.e. people with lived experience or clinicians) were only re-presented to the group which did not reach consensus.

Data analysis

There are no specific guidelines for determining consensus within a Delphi study.[231, 242, 243] For this study, we therefore determined consensus was achieved if at least 75% of

participants in each group (group 1: clinicians, group 2: people with lived experience) provided consistent ratings [232]:

1. Consensus: at least 75% of respondents in each group strongly agreed or agreed the modification was appropriate
2. Discarded: at least 75% of respondents in each group strongly disagreed or disagreed the modification was appropriate
3. Discordant: Items for which respondents could not agree. These items were retained and presented again in round 2.

Equal weighting was given across clinicians/researchers and people with lived experience. Optional open-ended responses were coded by one author (MS) to the pre-determined categories of content validity (comprehensibility, comprehensiveness, relevance) and feasibility, as defined by COSMIN.[244] Open-ended responses were specifically used to contextualise the discordant findings.

5.6 Results

Participant characteristics

Forty-four respondents completed round 1 of the survey (Table 5-1)), including clinicians (n=25), individuals with CP (n=9, mean age=34.48 years) and parents of children with CP (n=10, mean age of child=10 years). People with lived experience participants (group 2) were representative of all Gross Motor Function Classification System (GMFCS) levels and pain reporting abilities (able to self-report pain, unable to self-report pain and able to report pain with additional assistance) seen in CP. Thirty-seven respondents (84% retention) completed round 2 of the survey (Table 5-1), including clinicians (n=21), individuals with CP (n=8) and parents of children with CP (n=8).

Table 5-1 Descriptive statistics of Delphi study participants

Participant characteristic	**% (n)**	**% (n)**
	Round 1 (n=44)	Round 2 (n=37)
Clinicians	**56.8% (25)**	**56.8% (21)**
Orthopaedic Surgeon	4% (1)	0% (0)
Paediatrician	8% (2)	9.5% (2)

Participant characteristic	**% (n)**	**% (n)**
Physiotherapist	76% (19)	76.2% (16)
Psychologist	4% (1)	4.8% (1)
Speech Pathologist	4% (1)	4.8% (1)
Other	4% (1)	4.8% (1)
People with lived experience	**43.2% (19)**	**43.2% (16)**
Young people with CP	20.5% (9)	21.6% (8)
[age range]	[18-63]	[18-63]
Mean age years (SD)	34.89 (14.04)	35.5 (14.88)
8-18 years	0% (0)	n=0
18-30 years	55.5% (5)	50% (4)
> 30 years	44.5% (4)	50% (4)
Mobility level	n=9	n=8
GMFCS I	11.1% (1)	12.5% (1)
GMFCS II	66.7% (6)	75% (6)
GMFCS III	22.2% (2)	12.5% (1)
GMFCS IV	0% (0)	0% (0)
GMFCS V	0% (0)	0% (0)
Communication ability		
Able to report and describe pain without any additional assistance	100% (9)	100% (8)
Able to report and describe pain with the use of a communication device or other method	0% (0)	0% (0)
Unable to report and/or describe pain	0% (0)	0% (0)
Parents of children with CP	22.7% (10)	21.6% (8)
[child age range]	*[3-25]*	*[3-25]*
Mean age (SD)	*10 (7.5)*	*10 (7.5)*
Mobility Level (of child)	(n=10)	(n=8)
GMFCS I	20% (2)	12.5% (1)
GMFCS II	20% (2)	25% (2)
GMFCS III	20% (2)	12.5% (1)
GMFCS IV	20% (2)	25% (2)
GMFCS V	20% (2)	25% (2)
Communication ability (of child)		
Able to report and describe pain without any additional assistance	70% (7)	62.5% (5)
Able to report and describe pain with the use of a communication device or other method	10% (1)	12.5% (1)
Unable to report and/or describe pain	20% (2)	25% (2)

Round 1

Of the 37 suggestions presented to participants, 19 achieved consensus (Table 5-2, Table 5-3). Four suggestions reached consensus for the FOPQ-C-SF (Table 5-2), and fifteen suggestions for the mBPI (Table 5-3). No suggestions reached consensus to discard (>75% strongly disagree + disagree responses).

Table 5-2 Round 1 consensus results for FOPQ-C-SF modifications

Consensus rating: A = consensus reached for inclusion, B=no consensus reached, C=consensus reached for exclusion

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Examples fopq 1	Examples (item 2: Have a list of examples of what something terrible is e.g. trip over and hurt myself, have to go to hospital)	lived experience	52.6% (10)	31.6% (6)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	36% (9)	36% (9)	20% (5)	8% (2)	0% (0)	72	8	?
Examples fopq 2	Examples (item 9: Have some examples of equipment or therapy as part of I stop any activity if I start to hurt or my pain becomes worse e.g. using a standing frame, using a shower chair, using a wheelchair, going to physio)	lived experience	26.3% (5)	31.6% (6)	36.8% (7)	5.3% (1)	0% (0)	57.9	5.3	?
		clinician	28% (7)	36% (9)	16% (4)	16% (4)	4% (1)	64	20	?
Presentation fopq 1	Presentation: Make the font & response options larger and have bigger spacing between	lived experience	52.6% (10)	31.6% (6)	10.5% (2)	5.3% (1)	0% (0)	84.2	5.3	+
		clinician	40% (10)	52% (13)	8% (2)	0% (0)	0% (0)	92	0	+
Presentation fopq 2	Presentation: Have a version with only one question/item per page (including a picture, along with the response scale)	lived experience	31.6% (6)	36.8% (7)	21.1% (4)	10.5% (2)	0% (0)	68.4	10.5	?
		clinician	24% (6)	36% (9)	28% (7)	8% (2)	4% (1)	60	12	?
Proxy report fopq	Parent report: Do not include a parent report option for the Fear of Pain Questionnaire as it is too individual/personal and cannot be answered accurately by a parent	lived experience	21.1% (4)	21.1% (4)	26.3% (5)	26.3% (5)	5.3% (1)	42.2	31.6	?
		clinician	4% (1)	16% (4)	36% (9)	40% (10)	4% (1)	20	44	?
Response option fopq 1	Responses: Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	lived experience	36.8% (7)	31.6% (6)	21.1% (4)	10.5% (2)	0% (0)	68.4	10.5	?
		clinician	12% (3)	24% (6)	48% (12)	16% (4)	0% (0)	36	16	?
Response option fopq 2	Responses: Add visual symbols as well as numbers e.g. two thumbs up, one thumb up,	lived experience	36.8% (7)	47.4% (9)	10.5% (2)	5.3% (1)	0% (0)	84.2	5.3	+
		clinician	24% (6)	40% (10)	20% (5)	16% (4)	0% (0)	64	16	?

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
	thumbs in the middle, one thumb down, two thumbs down									
Visuals fopq 1	Visuals: Add pictures to each item e.g. feelings of pain are scary for me has a picture of someone looking scared	lived experience	36.8% (7)	42.1% (8)	21.1% (4)	0% (0)	0% (0)	78.9	0	+
		clinician	28% (7)	40% (10)	24% (6)	8% (2)	0% (0)	68	8	?
Wording fopq 1	Wording change (item 1: Remove the word normal Changes to I cant do all the things other people do because its so easy to hurt my body)	lived experience	78.9% (15)	15.8% (3)	0% (0)	5.3% (1)	0% (0)	94.7	5.3	+
		clinician	76% (19)	24% (6)	0% (0)	0% (0)	0% (0)	100	0	+
Wording fopq 2	Wording change (item 4: Change I cancel plans to I don't want to do things or I don't want to go to things)	lived experience	57.9% (11)	26.3% (5)	10.5% (2)	0% (0)	5.3% (1)	84.2	5.3	+
		clinician	52% (13)	28% (7)	8% (2)	12% (3)	0% (0)	80	12	+
Wording fopq 3	Wording change (items 4,7,8,10: Change words which assume autonomy to I wish I could or I want to e.g. Change I do not go to school because it makes my pain worse to I don't want to go to school because it makes my pain worse)	lived experience	52.6% (10)	36.8% (7)	5.3% (1)	5.3% (1)	0% (0)	89.4	5.3	+
		clinician	48% (12)	40% (10)	8% (2)	4% (1)	0% (0)	88	4	+

Consensus rating: + consensus reached for inclusion, ? no consensus reached, - consensus reached for exclusion

Table 5-3 Round 1 consensus results for mBPI modifications

Consensus rating: A = consensus reached for inclusion, B=no consensus reached, C=consensus reached for exclusion

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Addition mbpi 1	Wording addition (item 4: school/work to school/work/day activities)	lived experience	42.1% (8)	36.8% (7)	15.8% (3)	5.3% (1)	0% (0)	78.9	5.3	+
		clinician	60% (15)	32% (8)	0% (0)	8% (2)	0% (0)	92	8	+
Addition mbpi 2	Item addition: add a blank space to record a favourite activity and then rate how pain gets in the way of that	lived experience	36.8% (7)	42.1% (8)	15.8% (3)	0% (0)	5.3% (1)	78.9	5.3	+
		clinician	56% (14)	40% (10)	4% (1)	0% (0)	0% (0)	96	0	+
Addition mbpi 3	Addition: Add a comments section at the bottom to answer is there a time when you dont have pain? or is there something you do that makes your pain better?	lived experience	47.4% (9)	36.8% (7)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	48% (12)	40% (10)	8% (2)	4% (1)	0% (0)	88	4	+
Examples mbpi 1	Examples: Add examples/descriptions to each iteme.g. recreational activities = playing games with friends, going to the movies, swimming, listening to music	lived experience	57.9% (11)	36.8% (7)	5.3% (1)	0% (0)	0% (0)	94.7	0	+
		clinician	48% (12)	44% (11)	4% (1)	4% (1)	0% (0)	92	4	+
Examples mbpi 2	Examples: Add optional examples of equipment/assistive technology e.g. general activity (item 1 - include examples such as getting around in my walker/wheelchair/crutches)	lived experience	47.4% (9)	42.1% (8)	10.5% (2)	0% (0)	0% (0)	89.5	0	+
		clinician	36% (9)	44% (11)	16% (4)	4% (1)	0% (0)	80	4	+
Presentation mbpi 1	Presentation: Make the font larger and have bigger spacing	lived experience	52.6% (10)	36.8% (7)	5.3% (1)	5.3% (1)	0% (0)	89.4	5.3	+
		clinician	44% (11)	44% (11)	12% (3)	0% (0)	0% (0)	88	0	+

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Presentation mbpi 2	Presentation: Alternate row colours so it is easier to see	lived experience	26.3% (5)	36.8% (7)	21.1% (4)	15.8% (3)	0% (0)	63.1	15.8	?
		clinician	36% (9)	28% (7)	24% (6)	12% (3)	0% (0)	64	12	?
Presentation mbpi 3	Presentation: Have a version with only one question/item per page (including a picture, along with the response scale)	lived experience	26.3% (5)	42.1% (8)	21.1% (4)	10.5% (2)	0% (0)	68.4	10.5	?
		clinician	20% (5)	32% (8)	36% (9)	8% (2)	4% (1)	52	12	?
Proxy report mbpi	Parent report: A parent report option is appropriate for the modified brief pain inventory questions about how pain gets in the way of life	lived experience	47.4% (9)	36.8% (7)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	44% (11)	44% (11)	8% (2)	4% (1)	0% (0)	88	4	+
Response option mbpi 1	Responses: Have a version with less response options (i.e. reducing from 0-10 to 0-5)	lived experience	52.6% (10)	15.8% (3)	21.1% (4)	10.5% (2)	0% (0)	68.4	10.5	?
		clinician	32% (8)	44% (11)	12% (3)	12% (3)	0% (0)	76	12	+
Response option mbpi 2	Responses: Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	lived experience	36.8% (7)	36.8% (7)	5.3% (1)	21.1% (4)	0% (0)	73.6	21.1	?
		clinician	12% (3)	12% (3)	52% (13)	20% (5)	4% (1)	24	24	?
Response option mbpi 3	Responses: Add visual symbols as well as numbers e.g. happy face, okay face, unhappy face	lived experience	36.8% (7)	47.4% (9)	10.5% (2)	5.3% (1)	0% (0)	84.2	5.3	+
		clinician	28% (7)	36% (9)	20% (5)	16% (4)	0% (0)	64	16	?
Response option mbpi 4	Responses: Add descriptions along the response option scale e.g. on a 0-5 scale: 0 = not at all, 2 = a fair bit, 5 = a lot	lived experience	36.8% (7)	57.9% (11)	5.3% (1)	0% (0)	0% (0)	94.7	0	+
		clinician	36% (9)	48% (12)	12% (3)	4% (1)	0% (0)	84	4	+
	Responses: Add I don't know or I don't understand as a response option.	lived experience	73.7% (14)	26.3% (5)	0% (0)	0% (0)	0% (0)	100	0	+

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Response option mbpi 5	This is a prompt for people who use communication devices to be able to communicate that they need more explanation to answer the question	clinician	48% (12)	40% (10)	4% (1)	8% (2)	0% (0)	88	8	+
Visuals mbpi 1	Visuals: Add pictures to each item e.g. taking care of daily needs (item 6 has pictures of showering, getting dressed e.g. recreational activities (item 5 has pictures of outdoor activities, swimming and sport)	lived experience	52.6% (10)	36.8% (7)	5.3% (1)	5.3% (1)	0% (0)	89.4	5.3	+
		clinician	36% (9)	60% (15)	4% (1)	0% (0)	0% (0)	96	0	+
Visuals mbpi 2	Visuals: Include pictures of people using mobility aids and communication devices e.g. recreational activities (item 5 - has pictures of people with playing games but may be using a mobility aid e.g. communication with others (item 9 has pictures of a person using a high or low tech communication device))	lived experience	47.4% (9)	36.8% (7)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	48% (12)	52% (13)	0% (0)	0% (0)	0% (0)	100	0	+
Wording mbpi 1	Wording change: interferes changes to gets in the way	lived experience	42.1% (8)	42.1% (8)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	28% (7)	48% (12)	8% (2)	16% (4)	0% (0)	76	16	+
Wording mbpi 10	Wording change (item 11: social activities to spending time with friends and family)	lived experience	31.6% (6)	42.1% (8)	21.1% (4)	5.3% (1)	0% (0)	73.7	5.3	?
		clinician	44% (11)	32% (8)	16% (4)	8% (2)	0% (0)	76	8	+
Wording mbpi 2	Wording change (item 1: general activity to getting around)	lived experience	36.8% (7)	21.1% (4)	36.8% (7)	5.3% (1)	0% (0)	57.9	5.3	?
		clinician	36% (9)	32% (8)	8% (2)	20% (5)	4% (1)	68	24	?
Wording mbpi 3	Wording change (item 3: mood to feelings)	lived experience	31.6% (6)	26.3% (5)	26.3% (5)	15.8% (3)	0% (0)	57.9	15.8	?
		clinician	52% (13)	20% (5)	4% (1)	24% (6)	0% (0)	72	24	?

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Wording mbpi 4	Wording change (item 5: recreational activity to play or things I do for fun)	lived experience	42.1% (8)	47.4% (9)	10.5% (2)	0% (0)	0% (0)	89.5	0	+
		clinician	48% (12)	32% (8)	0% (0)	20% (5)	0% (0)	80	20	+
Wording mbpi 5	Wording change (item 6: taking care of daily needs to looking after myself)	lived experience	47.4% (9)	21.1% (4)	26.3% (5)	5.3% (1)	0% (0)	68.5	5.3	?
		clinician	36% (9)	40% (10)	16% (4)	8% (2)	0% (0)	76	8	+
Wording mbpi 6	Wording change (item 7: learning new information or skills to learning new things or concentrating)	lived experience	31.6% (6)	52.6% (10)	15.8% (3)	0% (0)	0% (0)	84.2	0	+
		clinician	36% (9)	44% (11)	4% (1)	16% (4)	0% (0)	80	16	+
Wording mbpi 7	Wording change (item 8: relations with others to friendships or getting along with others)	lived experience	57.9% (11)	26.3% (5)	10.5% (2)	5.3% (1)	0% (0)	84.2	5.3	+
		clinician	56% (14)	40% (10)	0% (0)	4% (1)	0% (0)	96	4	+
Wording mbpi 8	Wording change (item 9: communication with others to tell people what I want to tell them)	lived experience	31.6% (6)	21.1% (4)	26.3% (5)	21.1% (4)	0% (0)	52.7	21.1	?
		clinician	36% (9)	24% (6)	8% (2)	28% (7)	4% (1)	60	32	?
Wording mbpi 9	Wording change (item 10: enjoyment of life to having fun)	lived experience	31.6% (6)	36.8% (7)	31.6% (6)	0% (0)	0% (0)	68.4	0	?
		clinician	44% (11)	24% (6)	20% (5)	12% (3)	0% (0)	68	12	?

Consensus rating: + consensus reached for inclusion, ? no consensus reached, - consensus reached for exclusion

Round 2

Following round 2, a further nine changes reached consensus for inclusion (total 28/37); three changes to the FOPQ-C-SF (Table 5-4) and six to the mBPI (Table 5-5). No changes reached consensus to discard, and nine changes remained discordant. Table 5-6 and Table 5-7 detail the changes that reached consensus and those that were discordant.

Following round 2, a further nine changes reached consensus for inclusion (total 28/37); three changes to the FOPQ-C-SF (Table 5-4) and six to the mBPI (Table 5-5). No changes reached consensus to discard, and nine changes remained discordant. Table 5-6 and Table 5-7 detail the changes that reached consensus and those that were discordant.

Table 5-4 Round 2 consensus results for FOPQ-C-SF modifications

Suggestion		Experience	Strongly Agree %(n)	Agree %(n)	Neutral %(n)	Disagree %(n)	Strongly Disagree %(n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Examples fopq 1	Examples (item 2: Have a list of examples of what something terrible is e.g. trip over and hurt myself, have to go to hospital)	clinician	38.1% (8)	38.1% (8)	4.8% (1)	14.3% (3)	4.8% (1)	76.2	19.1	+
Examples fopq 2	Examples (item 9: Have some examples of equipment or therapy as part of I stop any activity if I start to hurt or my pain becomes worse e.g. using a standing frame, using a shower chair, using a wheelchair, going to physio)	lived experience	18.8% (3)	50% (8)	31.2% (5)	0% (0)	0% (0)	68.8	0	?
		clinician	23.8% (5)	28.6% (6)	38.1% (8)	4.8% (1)	4.8% (1)	52.4	9.6	?
Presentation fopq 2	Presentation: Have a version with only one question/item per page (including a picture, along with the response scale)	lived experience	6.2% (1)	62.5% (10)	18.8% (3)	6.2% (1)	6.2% (1)	68.7	12.4	?
		clinician	0% (0)	57.1% (12)	23.8% (5)	14.3% (3)	4.8% (1)	57.1	19.1	?
Proxy report fopq	Parent report: Do not include a parent report option for the Fear of Pain Questionnaire as it is too individual/personal and cannot be answered accurately by a parent	lived experience	12.5% (2)	6.2% (1)	37.5% (6)	31.2% (5)	12.5% (2)	18.7	43.7	?
		clinician	0% (0)	14.3% (3)	19% (4)	52.4% (11)	14.3% (3)	14.3	66.7	?
Response option fopq 1	Responses: Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	lived experience	12.5% (2)	50% (8)	25% (4)	12.5% (2)	0% (0)	62.5	12.5	?
		clinician	9.5% (2)	57.1% (12)	28.6% (6)	4.8% (1)	0% (0)	66.6	4.8	?
Response option fopq 2	Responses: Add visual symbols as well as numbers e.g. thumbs up/thumbs down	clinician	9.5% (2)	66.7% (14)	23.8% (5)	0% (0)	0% (0)	76.2	0	+
Visuals fopq 1	Visuals: Add pictures to each item e.g. feelings of pain are scary for me has a picture of someone looking scared	clinician	9.5% (2)	76.2% (16)	14.3% (3)	0% (0)	0% (0)	85.7	0	+

Consensus rating: + consensus reached for inclusion, ? no consensus reached, - consensus reached for exclusion

Table 5-5: Round 2 consensus results for mBPI modifications

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Presentation mbpi 2	Presentation: Alternate row colours so it is easier to see	lived experience	25% (4)	56.2% (9)	6.2% (1)	6.2% (1)	6.2% (1)	81.2	12.4	+
		clinician	38.1% (8)	52.4% (11)	4.8% (1)	4.8% (1)	0% (0)	90.5	4.8	+
Presentation mbpi 3	Presentation: Have a version with only one question/item per page (including a picture, along with the response scale)	lived experience	6.2% (1)	50% (8)	31.2% (5)	6.2% (1)	6.2% (1)	56.2	12.4	?
		clinician	0% (0)	57.1% (12)	28.6% (6)	9.5% (2)	4.8% (1)	57.1	14.3	?
Response option mbpi 1	Responses: Have a version with less response options (i.e. reducing from 0-10 to 0-5)	lived experience	37.5% (6)	37.5% (6)	18.8% (3)	0% (0)	6.2% (1)	75	6.2	+
Response option mbpi 2	Responses: Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	lived experience	12.5% (2)	37.5% (6)	37.5% (6)	6.2% (1)	6.2% (1)	50	12.4	?
		clinician	14.3% (3)	47.6% (10)	33.3% (7)	4.8% (1)	0% (0)	61.9	4.8	?
Response option mbpi 3	Responses: Add visual symbols as well as numbers e.g. happy face, okay face, unhappy face	clinician	4.8% (1)	85.7% (18)	4.8% (1)	0% (0)	4.8% (1)	90.5	4.8	+
Wording mbpi 10	Wording change (item 11: social activities to spending time with friends and family)	lived experience	43.8% (7)	43.8% (7)	6.2% (1)	0% (0)	6.2% (1)	87.6	6.2	+
Wording mbpi 2	Wording change (item 1: general activity to getting around)	lived experience	25% (4)	37.5% (6)	25% (4)	0% (0)	12.5% (2)	62.5	12.5	?
		clinician	28.6% (6)	52.4% (11)	9.5% (2)	9.5% (2)	0% (0)	81	9.5	+
Wording mbpi 3	Wording change (item 3: mood to feelings)	lived experience	56.2% (9)	25% (4)	12.5% (2)	6.2% (1)	0% (0)	81.2	6.2	+
		clinician	23.8% (5)	42.9% (9)	14.3% (3)	19% (4)	0% (0)	66.7	19	?

Suggestion		Experience	Strongly Agree % (n)	Agree % (n)	Neutral % (n)	Disagree % (n)	Strongly Disagree % (n)	Agree + Strongly Agree (%)	Disagree + Strongly Disagree (%)	Consensus Rating
Wording mbpi 5	Wording change (item 6: taking care of daily needs to looking after myself)	lived experience	62.5% (10)	25% (4)	6.2% (1)	0% (0)	6.2% (1)	87.5	6.2	+
Wording mbpi 8	Wording change (item 9: communication with others to tell people what I want to tell them)	lived experience	31.2% (5)	31.2% (5)	25% (4)	6.2% (1)	6.2% (1)	62.4	12.4	?
		clinician	38.1% (8)	38.1% (8)	4.8% (1)	9.5% (2)	9.5% (2)	76.2	19	+
Wording mbpi 9	Wording change (item 10: enjoyment of life to having fun)	lived experience	43.8% (7)	37.5% (6)	6.2% (1)	0% (0)	12.5% (2)	81.3	12.5	+
		clinician	23.8% (5)	57.1% (12)	14.3% (3)	0% (0)	4.8% (1)	80.9	4.8	+

Consensus rating: + consensus reached for inclusion, ? no consensus reached, - consensus reached for exclusion

Table 5-6 Results from round 2 – consensus changes to the mBPI and FOPQ-C-SF

Suggestions which reached consensus to include *>75% clinicians and >75% people with lived experience agreed or strongly agreed with the suggested change*	
Fear of Pain Questionnaire	
Change type	Change type
Presentation:	Make the font & response options larger and have bigger spacing between
Wording change (item 1):	Remove the word "normal" Changes to: "I can't do all the things other people do because it's so easy to hurt my body"
Wording change (items 4, 7, 8, 10):	Change words which assume autonomy to I wish I could or I want to e.g. Change I do not go to school because it makes my pain worse to I don't want to go to school because it makes my pain worse)
Wording change (item 4):	Change "I cancel plans" to "I don't want to do things" or "I don't want to go to things"
Examples (item 2)	Have a list of examples of what something terrible is e.g. trip over and hurt myself, have to go to hospital)
Response option	Add visual symbols as well as numbers e.g. two thumbs up, one thumb up, thumbs in the middle, one thumb down, two thumbs down
Visuals	Add pictures to each item e.g. feelings of pain are scary for me has a picture of someone looking scared
Modified Brief Pain Inventory	
Addition:	Add a comments section at the bottom to answer "is there a time when you don't have pain?" or "is there something you do that makes your pain better?"
Examples	Add examples/descriptions to each item e.g. recreational activities = *playing games with friends, going to the movies, swimming, listening to music*
Examples	Add optional examples of equipment/assistive technology *e.g. general activity (item 1) – Include examples such as getting around in my walker/wheelchair/crutches*
Item addition	Add a blank space to record a favourite activity and then rate how pain gets in the way of that
Presentation	Make the font larger and have bigger spacing
Presentation	Alternate row colours so it is easier to see
Parent report	A parent report option is appropriate for the modified brief pain inventory questions about how pain gets in the way of life
Responses	Add descriptions along the response option scale e.g. *on a 0-5 scale: 0 = not at all, 2 = a fair bit, 5 = a lot*
Responses	Have a version with less response options (i.e. reducing from 0-10 to 0-5)
Responses	Add "I don't know" as a response option
Responses	Add visual symbols as well as numbers e.g. happy face, okay face, unhappy face
Visuals	Add pictures to each item *e.g. taking care of daily needs (item 6) has pictures of showering, getting dressed* *e.g. recreational activities (item 5) has pictures of outdoor activities, swimming and sport*
Visuals	Include pictures of people using mobility aids and communication devices *e.g. recreational activities (item 5) - has pictures of people with playing games but may be using a mobility aid* *e.g. communication with others (item 9) has pictures of a person using a high or low tech communication device*
Wording change	"Interferes" changes to "gets in the way"
Wording addition (item 4)	Change "School/work" to "school/work/day activities"

Suggestions which reached consensus to include *>75% clinicians and >75% people with lived experience agreed or strongly agreed with the suggested change*	
Wording change (item 5)	"Recreational activity" to "play" or "things I do for fun"
Wording change (item 6)	"Taking care of daily needs" to "looking after myself "
Wording change (item 7)	"Learning new information or skills" to "learning new things" or "concentrating"
Wording change (item 8)	"Relations with others" to "friendships" or "getting along with others"
Wording change (item 10)	"Enjoyment of life" to "having fun"
Wording change (item 11)	"Social activities" to "spending time with friends and family"

Table 5-7 Results from round 2: Discordant suggested changes to the mBPI and FOPQ-C-SF

Fear of Pain Questionnaire					
Change type	**Change**	**Clinicians** (agree/strongly agree %)	**People with lived experience** (agree/strongly agree %)	**Open-ended response category**	**Summary**
Examples (item 9)	Have some examples of equipment or therapy as part of I stop any activity if I start to hurt or my pain becomes worse e.g. using a standing frame, using a shower chair, using a wheelchair, going to physio	52%	69%	Relevance Comprehens-iveness	Implies the activities are only related to therapy Could bias the answers those with less physical disability (i.e. those who don't use equipment)
Presentation	Have a version with only one question/item per page (including a picture, along with the response scale	57%	69%	Feasibility	Challenging in clinical settings due to multiple pieces of paper, would be suitable as an electronic version
Proxy report	Parent report: Do not include a parent report option for the Fear of Pain Questionnaire as it is too individual/personal and cannot be answered accurately by a parent	Agree/strongly agree: 14% Disagree/strongly disagree: (67%)	Agree/strongly agree 19% Disagree/strongly disagree: (43%)	Feasibility Relevance	Useful to know the **parent's own** beliefs around pain (rather than the parent's perspective of the child's beliefs about pain). Sometimes the child won't be able to report at all, and in this case parent information could be useful.
Response option	Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	67%	63%	Feasibility Comprehens-ibility	Increased visual complexity for those with visual impairment Colours could influence the responses as they can have an emotional overlay

Modified Brief Pain Inventory					
Change type	**Change**	**Clinicians**	**People with lived experience**	**Open-ended response category**	**Summary**
Presentation	Have a version with only one question/item per page (including a picture, along with the response scale	57%	56%	Feasibility	Challenging in clinical settings due to multiple pieces of paper
Responses	Add colour background to the response options e.g. changes from green to yellow to red as the score increases e.g. changes from lighter shade to darker shade of the same colour as the score increases	62%	50%	Feasibility Comprehens-ibility	Increased visual complexity for those with visual impairment Colours could influence the responses as they can have an emotional overlay
Wording change (item 1)	"General activity" to "getting around"	81%	63%	Comprehens-iveness Relevance	General activity is more than just 'getting around'. It should also encompass other activities that are not mobility dependent
Wording change (item 3)	"Mood" to "feelings"	67%	81%	Comprehens-ibility	Mood and feelings can be interpreted differently. Feelings is too broad, mood more clearly relates to 'low mood' (i.e. pain, anxiety, depression)
Wording change (item 9)	"Communication with others" to "tell people what I want to tell them"	76%	62%	Comprehens-iveness	Communication is more than just telling, it also needs to include listening, understanding and being understood.

Agreement between groups

In round 1, four suggested changes reached consensus for people with lived experience but not for clinicians (Table 5-2). All four of these changes reached consensus with clinicians in round 2. Three suggested changes in round 1 reached consensus for clinicians but not people with lived experience (Table 5-4); again all of these changes reached consensus with people with lived experience in round 2. At the end of round 2, only three suggested changes, all related to the mBPI, had reached consensus with one group only (Table 5-7).

5.7 Discussion

This study highlighted the importance of modifying assessment tools to improve the validity and feasibility of the tools for use in young people with CP and chronic pain. Clinicians and people with lived experience gained consensus on modifications to the FOPQ-C-SF and the mBPI to improve appropriateness and accessibility for young people with CP. These modifications are now suitable for implementation and pilot testing in children and young people with CP and varying communication and cognitive abilities.

The modifications to be implemented include wording adjustment, improvement of the visual presentation of the tool, use of visuals to aid understanding and a reduction in the number of response options. The desire to include visuals and/or symbols is unsurprising, with Rinde and colleagues (2023) reporting good success in engaging children with CP, complex communication needs and reduced cognitive abilities in discussion about their pain when using visual aids.[209] Similarly, reducing the number of response options has been shown to improve the reliability of self-reported pain intensity in typically developing three and four year olds.[245] Adding examples of equipment or assistive technology to mBPI items, for instance mobility (item 12 mBPI) = getting around in my walker/wheelchair/crutches, reached consensus. This is important with research conducted by Dubois and colleagues (2023) reporting standing frame use was more frequently associated with pain than any other care related acts experienced by children with CP.[74] The wording changes in the FOPQ-C-SF also recognise the reduced autonomy children and young people with disability may have as a result of mobility, cognition or communication limitations.[246]

Feasibility and content validity are both key in recommending a tool for use in clinical practice.[27] Feasibility is crucial, as it determines the uptake of a tool by people with lived experience and clinicians in clinical practice.[196] Content validity is considered the most important measurement property, as it determines whether a tool measures what it intends to measure, including relevance to the population of interest, relevance to the construct of interest, comprehensiveness and comprehensibility for the target population.[27] Our previous qualitative study investigated the content validity and feasibility of the original mBPI and FOPQ-C-SF, generating suggestions for improving these tools for children and young people with CP. Subsequently, this current study focused on selecting the most important modifications to enhance relevance, comprehensiveness, comprehensibility and feasibility for this population, without changing the construct of interest, ready for further testing. Unfortunately, we did not have any children with CP (<18 years) willing to participate in the Delphi process, although they were eligible to be included. While it is possible this may have skewed the modifications which did reach consensus, the diversity of adults with CP and parents of children with CP included was likely sufficient to overcome this. While this is a limitation of this study, we do not consider it a significant limitation in the overall adaptation of the tools as our previous research has thoroughly investigated both the relevance and comprehensiveness of the modified versions of the FOPQ-C-SF and mBPI through qualitative interviews with children with CP. We are also cognisant that comprehensibility requires further testing across a broader range of children with CP and cognitive and communication impairment; it was not possible to include the full breadth of cognitive and communication impairment in this study or our previous qualitative study due to the complexity of the original versions of the tools.

Two wording suggestions for the mBPI reached consensus with clinicians but not people with lived experience. Whilst clinicians suggested changing “general activity” to “getting around”, which essentially combined general activity and mobility into one item, a number of participants provided open ended responses indicating general activity is “more than just moving around” (Table 5-7). We therefore suggest the wording of “general activity” be further investigated with children and young people with CP. A further suggestion for the mBPI, changing “mood” to “feelings” reached consensus for people with lived experience but not clinicians. Open ended response comments from

clinicians suggested the term 'feelings' is much broader than "mood", and "feelings" are not necessarily linked with anxiety, depression and pain in the same way as "mood" (Table 5-7). We recommend this also needs further investigation.

One suggestion for this study was to have a version of each assessment tool listing one question or item per page. Although the three lived experience participants representing children with complex communication needs all agreed or strongly agreed with this suggestion, it did not reach consensus in either group. Clinician feedback indicated potential difficulty of implementing an assessment with multiple pages in clinical practice, however this feedback also suggested use of a digital device may make this feasible.[211] Despite this suggestion not reaching consensus, given the importance of this modification for AAC users, we recommend it is further considered and tested in children with complex communication needs.

A number of the modifications to the tools related to comprehensibility, including simplifying wording, using visuals to assist understanding and simplifying response categories. Most paediatric patient reported outcome measures (PROMs) advise self-report questionnaires are suitable for children aged 8 years and above.[247] This age recommendation is based on cognitive ability. However, as 1 in 2 children with CP have an intellectual disability[57], setting the standard for self-reporting at a cognitive age of 8 years potentially excludes a large proportion of the population from ever accessing self-report measures. Given the higher prevalence of chronic pain in the CP population[3], consideration needs to be given as to how best to adapt PROMs to be more accessible to children with cognitive abilities below 8 years of age.[248] This is especially important considering the presence of both motor and cognitive disabilities, limited communication skills and frequent care interventions place these individuals at a high risk of experiencing repeated induced pain.[74] Emmott and colleagues (2017) have been considering how to improve access to self-reported pain intensity measures in typically developing children as young as 3 years of age.[245] Despite the challenges in eliciting self-report in young children, some degree of self-report is still preferred given the reduced accuracy of proxy report.[120, 210, 245] This can be performed in conjunction with parent proxy report where the accuracy of the child's self-report is unclear.

The need for holistic and accessible pain assessment for children with developmental disabilities like CP is increasingly recognised.[112] Further, ensuring all twelve core pain

domains for CP[233] are assessed requires clinicians to think more broadly than just pain intensity.[22] The FOPQ-C-SF and mBPI can both be used to assess the impact of pain on emotional functioning. The mBPI can be used as a multidimensional tool to contribute to assessment of this domain, and the FOPQ-C-SF provides assessment of pain related-fear and avoidance.[236] Routine assessment of the impact of pain on emotional functioning may improve access to currently under-utilised best practice multidisciplinary interventions for children and young people with CP.[34]

Our study had a number of strengths. Participants in the study had close to equal representation of clinicians and people with lived experience, and lived experience participants were representative of the spectrum of abilities seen in the CP population. We also used a modified two stage Delphi design which likely improved participation in round 2. A response rate of 84% was achieved in round 2, which is well above the recommended minimum response rate of 70% for subsequent rounds of a Delphi study.[249] It is possible the summary of round one results influenced participants' responses in round two. However, this is standard Delphi procedure.[232] The results for the lived experience and clinician groups were presented separately to acknowledge group effects and minimise the influence of clinicians on people with lived experience.[231] A study limitation was that although a range of clinician disciplines were represented, the majority of participants were physiotherapists. While this may have potentially skewed which modifications reached consensus, this was managed through assigning equal weighting to both people with lived experience and clinicians. Furthermore, as it is highly likely physiotherapists will be one of the larger discipline groups using the modified tools in clinical practice, we do not consider this a significant limitation. Finally, the survey included an optional open-ended comment box for each suggested change, which not all participants completed. As a result, the comments may not fully represent the entire sample. The open-ended comments were also only coded by one author. However, since these comments were used only to provide possible explanations and context for changes that did not reach consensus, this is not considered a significant limitation.

5.8 Conclusion

Consensus has been gained on modifications to the FOPQ-C-SF and mBPI to improve accessibility and self-report opportunity for all children and young people with CP. Future research will involve implementing these modifications and then pilot testing the tools across the communication, cognitive and motor ability spectrum of children and young people with CP. This will allow for these assessment tools to be used in clinical practice to increase access to best practice chronic pain interventions for children and young people with CP.

Chapter 6 – The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy (Study 4)

6.1 Preamble

Implementing modifications to two pain assessment tools

Chapter 5 reported the results of a consensus study to determine which modifications should be implemented to the FOPQ-C-SF and the mBPI for young people with CP. Consensus was reached on four modifications to the FOPQ-C-SF and 21 to the mBPI, including modifications to wording, response options, administration methods and visual presentation. One of the most important changes suggested for the mBPI, to enable self-report from AAC users and young people with cognitive impairment, was to reduce the number of response options from an 11-point to 5-point scale. Using fewer response options has also been found to be an effective strategy in improving the ability of preschool-age children (3-4 years of age) to self-report pain intensity.[245, 250, 251] The adaptations implemented to the content, layout and administration procedures of the tools were consistent with several features of cognitive accessibility, defined as "reducing the cognitive demands and/or supporting the cognitive processes to enable respondents to interpret and respond to the assessment items as intended".[248] A graphic designer was engaged to implement the recommended visual presentation modifications.

A key recommendation from both clinicians and people with lived experience in Chapter 4 was to consider alternative administration methods for people with diverse cognitive and communication abilities. For this reason, *Talking Mats® Ltd,* a UK-based visual communication social enterprise, were engaged to co-develop *Talking Mat* versions of the FOPQ-C-SF and mBPI. *Talking Mats®* is an evidence-based visual communication framework to promote understanding and expression of views for people with communication disability.[229]

This process resulted in the development of both a standard paper version and a *Talking Mat* version of the adapted mBPI and FOPQ-C-SF. Chapter 6 reports the findings of a mixed-methods study using these newly modified tools in a sample of children and young people with cerebral palsy. The study aim was to determine if the newly modified tools were comprehensible and feasible for use in children and young people with CP, and if any further changes to the tools were required.

6.2 Authorship declaration

This manuscript is published in *Quality of Life Research*.

Smith, M.G.[1,2,3], Gibson, R.J.[1], Schibani, M.[4], Russo, R.N.[4,5], Thirumanickam, A.[1], Harvey, A.R[6,7]. The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy: a multi-stakeholder consensus study. *Quality of Life Research* **34**, 2377–2392 (2025). https://doi.org/10.1007/s11136-025-03981-4

[1]School of Allied Health Science and Practice, University of Adelaide, Adelaide, Australia;
[2]Novita, Adelaide, Australia;
[3]Women's and Children's Hospital, Adelaide, Australia;
[4]Paediatric Rehabilitation Department, Women's and Children's Hospital, Adelaide, Australia;
[5]College of Medicine and Public Health, Flinders University, Adelaide, Australia;
[6]Neurodisability and Rehabilitation, Murdoch Children's Research Institute, Melbourne, Australia;
[7]Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia

Statement of Authorship

Title of Paper	The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy
Publication Status	X Published ☐ Accepted for Publication ☐ Submitted for Publication ☐ Unpublished and Unsubmitted work written in manuscript style
Publication Details	Smith, M.G., Gibson, R.J., Schibani, M., Russo, R.N., Thirumanickam, A., Harvey, A.R. The comprehensibility and feasibility of the modified Brief Pain Inventory and Fear of Pain Questionnaire adapted for children and young people with cerebral palsy. *Quality of Life Research*. (2025). https://doi.org/10.1007/s11136-025-03981-4

Principal Author

Name of Principal Author (Candidate)	Meredith Smith		
Contribution to the Paper	Completed data collection, analysis of all data, wrote manuscript and acted as corresponding author		
Overall percentage (%)	60%		
Certification:	This paper reports on original research I conducted during the period of my Higher Degree by Research candidature and is not subject to any obligations or contractual agreements with a third party that would constrain its inclusion in this thesis. I am the primary author of this paper.		
Signature		Date	6/1/25

Co-Author Contributions

By signing the Statement of Authorship, each author certifies that:

i. the candidate's stated contribution to the publication is accurate (as detailed above);

ii. permission is granted for the candidate in include the publication in the thesis; and

iii. the sum of all co-author contributions is equal to 100% less the candidate's stated contribution.

Name of Co-Author	5% Mathew Schibani		
Contribution to the Paper	Contributed to data collection and analysis, edited manuscript		
Signature		Date	6/1/25
Name of Co-Author	5% Abi Thirumanickam		
Contribution to the Paper	Contributed to data analysis, edited manuscript		
Signature		Date	22/1/25
Name of Co-Author	10% Adrienne Harvey		

Contribution to the Paper	Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	22/1/25
Name of Co-Author	10% Rachel Gibson		
Contribution to the Paper	Supervised development of work, edited manuscript		
Signature		Date	10/2/25
Name of Co-Author	10% Ray Russo		
Contribution to the Paper	Supervised development of work, edited manuscript		
Signature		Date	6/1/2025

6.3 Abstract

Purpose: To test the comprehensibility and feasibility of the modified brief pain inventory (mBPI) and Fear of Pain Questionnaire for Children short-form (FOPQ-C-SF) adapted for children and young people with cerebral palsy (CP), with diverse cognitive and communication abilities. Improving assessment of pain interference (mBPI) and pain-related fear (FOPQ-C-SF) in CP can enhance quality of life by increasing access to under-utilised interventions targeting pain-related physical disability and mental health.

Method: A convergent mixed methods approach was used. Twenty-two people (5-30 years) with CP completed the adapted mBPI and FOPQ-C-SF in a cognitive interview, administered by pen/paper or *TalkingMats®,* an evidence-based visual communication framework. Cognitive interviewing approaches were adapted to optimise participation and expression for diverse cognitive and communication abilities. Quantitative data were analysed to report tool administration times, overall completion rates and communication effectiveness. Qualitative data were analysed by content analysis to determine further changes required to the tools.

Results: Median administration times were 6.2 min (IQR=5.3-7.6) for mBPI and 4.1 min (IQR=2.7-4.9) for FOPQ-C-SF. All completed the mBPI. Three did not complete the FOPQ-C-SF due to fatigue, challenging behaviour and parent recommendation. Ten minor changes were identified.

Conclusion: The adapted mBPI and FOPQ-C-SF are likely comprehensible and feasible for children and young people with CP, including those with diverse cognitive and communication abilities. It is likely most children and young people with CP can effectively communicate responses to both tools. The adapted tools will now undergo further psychometric testing, prior to becoming freely available for clinical and research use.

6.4 Introduction

Children and young people with cerebral palsy (CP) experience higher rates of chronic pain than other paediatric populations, with reported prevalence up to 76% [3]. Noyek et al. (2023) identified pain intensity as the most common chronic pain domain assessed in young people with developmental disabilities [112]. However pain intensity contributes little to understanding chronic pain within the context of a person's life. Pain should instead be assessed within a biopsychosocial framework [21]. A challenge when applying a holistic chronic pain assessment framework is that many available tools are not validated in CP, or lack suitability across the spectrum of motor, communication and cognitive abilities in the CP population [24]. Cognitive impairment can make it challenging to meet the cognitive demands required of a self-report assessment, including attention to task, memory and judgement [248]. Practically, this includes understanding the item itself, relating the item to the individual's experience, understanding the response scale and making an evaluation of which response best represents the individual. For a child or young person with CP, this may be further compounded by the additional cognitive and motor demands of using augmentative and alternative communication (AAC), or fine motor limitations (i.e. for writing or circling the items). Despite these challenges, efforts to support individuals with diverse cognitive and communication abilities to self-report their pain should be prioritised [112]. Improved pain assessment has the potential to increase access to currently under-utilised best-practice chronic pain interventions, leading to better pain management and an overall improvement in quality of life [34].

Previous research by our team has identified two tools for potential use in CP [211]: the Modified Brief Pain Inventory (mBPI) [238], a modified version of the Brief Pain Inventory assessing pain interference in children and young people with neuromuscular disorders, and the Fear of Pain Questionnaire for Children – Short Form (FOPQ-C-SF) [235], which assesses pain-related fear and activity avoidance in children. Pain interference and pain-related fear/activity avoidance are key factors to consider when evaluating the impact of pain on quality of life [204]. Furthermore, these factors are often targets for chronic pain interventions, highlighting the importance of accurate assessment. For example, improving the assessment of pain-related fear could lead to better identification of children and young people with CP who may benefit from

psychological interventions, helping them to manage their chronic pain more effectively and, ultimately, improving their participation in activities meaningful to them.
Both tools have undergone evaluation of two components of content validity (relevance and comprehensiveness) to identify modifications to improve appropriateness and accessibility for children and young people with CP, which have since been completed [211]. The modifications implemented have sought to reduce cognitive demand and improve accessibility, including by simplifying wording, using alternative administration methods and adding visual symbols. Further investigation of comprehensibility was recommended, defined as 'ensuring tools are understood as intended by the population of interest' [240].

Therefore, the primary aim of this study was to evaluate the comprehensibility and feasibility of the newly adapted tools in children and young people with CP, encompassing a range of cognitive, communication and motor abilities. A secondary aim was to determine if further changes were required. We aimed to answer the following research questions:

1. Are the adapted assessment tools comprehensible for children and young people with CP, with diverse cognitive, communication and motor abilities?
2. Are the adapted assessment tools feasible for use with children and young people with CP, including those who use AAC?
3. Are further changes required to improve the comprehensibility and feasibility of the tools?

6.5 Methods

Study design

This study employed a convergent mixed methods approach[252], integrating prospective observational (quantitative) and qualitative descriptive components[226]. Both quantitative and qualitative data were concurrently collected and analysed: the quantitative to examine administration time and effectiveness of communication interactions, and the qualitative using modified cognitive interviewing to understand children and young people's perceptions. This approach allowed for a comprehensive analysis of the research topic, adhering to the consolidated criteria for reporting

qualitative research (COREQ)[213] and guided by the COnsensus-based Standards for Measurement Instruments (COSMIN)[240]. The study was approved by the Women's and Children's Health Network Human Research Ethics Committee (2022/HRE00154) and guided by a lived experience advisory group, including two young adults with CP and a parent of a child with CP, which was representative of the spectrum of motor, cognitive and communication ability seen in CP. The advisory group met bimonthly throughout the project and were reimbursed for their time. They provided guidance on all aspects of the project including the research questions, participant recruitment strategies, interview engagement strategies, piloting the interview guide, reviewing the qualitative coding schema and assisting with interpretation of findings.

Participants and setting

Children and young people were eligible for inclusion if they met the following criteria: a) diagnosis of CP, b) 5-30 years old, c) able to communicate a simple response by any method and d) experienced 'everyday pain', defined as 'pain which is recurrent or chronic with acute exacerbations' [34]. The advisory group recommended the age group inclusion criteria to 1) ensure the modifications made to the tools to enhance cognitive accessibility enabled understanding for a person with the cognitive ability of a five-year-old child; and 2) to extend the cut-off for young adulthood to 30 years, recognising this period may be extended for individuals with disability due to delayed transitions to adulthood social roles. It was expected that most participants would fall within levels I-III of the Communication Function Classification System (CFCS), meaning they would typically be able to communicate effectively with at least familiar partners, in order to meet inclusion criteria c). There were no specific exclusion criteria. Children and young people were recruited by online advertising and flyer distribution through Novita and the Women's and Children's Paediatric Rehabilitation Department. Maximum purposeful sampling was employed to facilitate inclusion of varied ages and abilities. Written informed consent was obtained from young adults without cognitive impairment, caregivers of children and caregivers of young adults with cognitive impairment. The ethics approved protocol contained guidelines to ensure children and young people understood the study (where possible) and/or to obtain assent.

Sample size

COSMIN consider testing content validity in a qualitative study of seven or more participants from the population of interest as 'very good', and four to six participants 'adequate' [240]. Given the heterogenous population, we chose to target 15-20 children and young people in a larger single round, rather than multiple smaller rounds. A similar approach was used by Kramer and colleagues, when testing the comprehensibility of a functional performance patient-reported outcome measure (PROM) [253]. We also chose to target at least 4 participants in each subgroup of interest (age group, motor ability, cognitive ability and communication ability) to ensure adequate representation.

Materials

Talking Mat versions of the mBPI and FOPQ-C-SF

One key recommendation from our previous work was to consider alternative administration methods for the tools [211]. *TalkingMats Ltd*, an evidence-based visual communication social enterprise was engaged to develop an alternative for each tool. *TalkingMats* has been previously used to support understanding and improve the expression of views of people with disability [229]. An iterative process including reviewing the modifications, reaching agreement on further minor modifications and selecting visual symbols was undertaken (MGS, ARH, RJG, RNR, *TalkingMats Ltd,* advisory group). This resulted in adapted versions of the mBPI and FOPQ-C-SF, in two formats: pen/paper and *TalkingMats*. The *TalkingMats* response scale contained three symbols, with each item able to be placed either under a symbol or in between the symbols, producing a 5-point scale consistent with the paper version. Typically a *TalkingMat* uses only positive wording for the response option on the left side of the mat, however we used a negative phrase to enable direct comparison between the paper and *TalkingMat* versions [229]. The pen/paper version also included identical response symbols, along with the same visual symbols for each item.

Effectiveness Framework of Functional Communication

The Effectiveness Framework of Functional Communication (EFFC) was used alongside cognitive interviewing to determine the young person's understanding of the tool and ability to provide accurate responses. The EFFC was chosen as it is a valid and reliable

tool assessing the effectiveness of a communication interaction and considers domains such as engagement and understanding of both the thinker and listener [229, 254].

Data collection

Children and young people were offered a choice of interview location; home, a community site or online. Interviews were 30-60 minutes duration and conducted by one of two interviewers (MGS, MS) between November 2023 and January 2024. All interviews were audio-visually recorded, transcribed verbatim and able to be ceased at any time. Interviews with children were conducted with a caregiver present, with some children and young people known to MS/MGS through clinical relationships. Children and young people were randomly assigned a *TalkingMats* or paper version of the mBPI and FOPQ, unless they required a specific version for both as determined by the caregiver and interviewer (Table 6-1). Caregivers and support people were requested to provide communication support only, focusing on eliciting the participant's voice. Caregivers were asked to withhold their perceptions and views on the questions until the end of the interview, with interviewers reiterating this during the interview as necessary. Interviewers recorded field notes both during, and immediately following, the interview (template in Appendix E1). Approaches from the literature were combined to develop an interview guide (Appendix E2) with three main stages (Table 6-2) [253, 255-257].

Table 6-1 Strategies implemented to facilitate participation from young people with varying cognitive and communication abilities at different stages of the project

Recruitment	**Prior to the interview**	**During the interview**	**After the interview**
Asking clinicians to invite young people to participate rather than relying solely on online advertising	Discussing communication methods and approaches for the young person with the caregiver/communication support person prior to the interview	Allowing a support person to be present during the interview	Allowing asynchronous participation- option to leave symbols with the young person after the interview for further consideration
Specifically inviting AAC users on the flyer	Caregivers and the interviewer were able to select the most appropriate version of the tools for testing (and able to switch to a different administration method during the interview if needed)	Visual support	
	Allowing asynchronous participation - including the option to view symbols in advance	Advising support people of the importance of eliciting the participant's voice in the interview rather than that of the support person	
		Allowing longer interview times for AAC users	
		Video recording interviews to capture non-verbal communication	

Table 6-2 Interview guide stages

Further specific information is provided in Appendix *E2*

Stage 1	Observation of the tool being completed, including asking the young person to 'think out loud' if possible
Stage 2	Probing questions focused on particular items, response options, symbols/pictures, examples, clarifying interpretation
Stage 3	Overall experience of the tool, supported by a visual evaluation in the form of a simple 3-point Likert scale. If applicable, parents were also engaged in this stage and asked to provide feedback on their child's engagement with the process, understanding and preference for administration method

The interview guide was tested by the advisory group, who were asked to reflect on their experience as a young person/their own child's experience and suggest changes. As the *TalkingMat* and pen/paper versions use identical questions, symbols and response options, the administration method was not expected to affect comprehensibility. However, it was anticipated to influence feasibility, including the participant's engagement in the cognitive interview itself. For this reason, the advisory group recommended specifying the administration method (*TalkingMat* vs paper) could be changed during the interview if it was not meeting the needs of the interviewee. Furthermore, interview methods were adapted to each person's abilities, using evidence-based strategies to optimise participation and expression for those with varying cognitive abilities [209, 258] and those who used AAC [259] (Table 6-1).

Data analysis

The EFFC was scored independently by two of three authors for each young person (MGS, ARH, MS). An overall score was provided for each tool completed by the young person. Following initial scoring, meetings were held to reach consensus on each item and overall score. Where consensus was unable to be reached, a third author was involved. Consensus EFFC scores were totalled for each tool and version. A score of ≥75% (21) was considered 'effective' communication as per Murphy et al. (2010) [254].

Each item on the EFFC is scored on a 0-4 scale (0=never/none, 4=always). For example, a score of 75% could be obtained by scoring 'often=3' for each of the six domains (engagement, thinker's understanding of the issue for discussion, listener's understanding of thinker's views, thinker on track, symmetry, real time and listener's satisfaction). Quantitative interview data was descriptively analysed to report tool administration duration, overall completion rates, missing items and consensus EFFC scores.

Qualitative data, including videos, transcripts, interview notes and completed questionnaires were analysed by a two-stage content analysis (Table 6-3) [226, 260]. This adapted approach ensured research questions could be answered practically and specifically, while still allowing for deeper exploration [260]. All data was analysed independently by two of three authors for each young person (MS, ARH, MS). Data from children and young people using AAC devices were also analysed by an experienced speech pathologist (AT). Transcripts and quotes were offered for review. NViVO 12.0plus software (QSR International, Melbourne, Australia) was used to facilitate analysis. The final coding schema was triangulated with the authors, advisory group and EFFC scores. This was an iterative process conducted by the authors and the advisory group, resulting in the final coding framework (Table 6-6) and item matrix table (Appendix E3). The item matrix table was used to identify items with unintended interpretation and track changes to each specific item in each tool (Appendix E3). All suggested changes were reviewed by the authors and advisory group to reach consensus on the final version (Table 6-6, Appendix E3).

Table 6-3 Stages of qualitative content analysis

Stage 1 - deductive	Stage 2 - inductive
1. Initial familiarisation with data • Watching video recordings • Reviewing transcripts/interview notes • Reviewing final questionnaires	1. Sub-category classification – including potential explanations for 'unintended interpretation' comprehensibility data
2. Coding data to pre-determined big picture coding categories related to the research questions (comprehensibility, feasibility, suggested changes, other)	2. Consensus determination for sub-category definition/description (ARH, MGS, MS)
3. Coding comprehensibility data as 'intended' or 'unintended' interpretation in relation to item (wording and symbol), instructions and recall period	4. Continued sub-category coding
	5. Review of the coding schema and all suggested changes (all authors and advisory group), including triangulation with EFFC scores to determine possible reasons for unintended interpretation and feasibility issues
	6. Iterative discussion with advisory group and research team to finalise the coding schema, item matrix table and the changes to be implemented

Researcher characteristics and reflexivity

Content analysis recognises the researcher's perspectives and prior experiences may influence data analysis [226]. MGS is a female PhD student and physiotherapist, and MS is a male occupational therapist, both with >9 years' experience working with children and young adults with CP. AT is an experienced qualitative researcher and speech pathologist working with children who use AAC. ARH, an experienced disability researcher and physiotherapist; RR, a paediatric rehabilitation medical specialist and researcher, and RJG, an experienced researcher in both quantitative and clinical studies, supervised the research. MGS, MS and ARH all underwent formal training in *TalkingMats*.

6.6 Results

Twenty-two children and young people with CP and diverse motor, communication and cognitive abilities participated (Table 6-4, Appendix E4). Qualitative data were coded to two main categories and seven subcategories (Table 6-5). The quantitative and qualitative results have been integrated and are presented within the two main categories of feasibility and comprehensibility. A full list of quotes is provided in Appendix E5.

Table 6-4 Participant demographics

Participant characteristic	**% (n)**	**Mean (SD)**
Age	n=22	14.91 years (6.21 years) Age range: 5-29 years
5-12 years	36.4% (8)	
13-17 years	31.8% (7)	
18-30 years	31.8% (7)	
Sex		
Male	54.5% (12)	
Female	45.5% (10)	
Gross Motor Function Classification System (GMFCS) level		
I	13.6% (3)	
II	45.5% (10)	
III	22.7% (5)	
IV	13.6% (3)	
V	4.5% (1)	
Manual Ability Classification System (MACS) level		
I	31.8% (7)	
II	36.4% (8)	
III	4.5% (1)	
IV	22.7% (5)	
V	4.5% (1)	
Communication Function Classification System (CFCS) level		
I	40.9% (9)	
II	40.9% (9)	
III	13.6% (3)	
IV	4.5% (1)	

Participant characteristic	% (n)	Mean (SD)
Communication ability in relation to pain		
Able to report and describe pain without any additional assistance	81.8% (18)	
Able to report and describe pain with the use of a communication device or other method	18.2% (4)	
Parent-reported cognitive impairment (CI)^		
Likely	50% (11)	
mild	9.1% (2)	
moderate	31.8% (7)	
unsure	4.5% (1)	
Unlikely	50% (11)	
Formal cognitive assessment completed		
No	72.7% (16)	
Yes	27.3% (6)	
School type	**(n=15)**	
Mainstream without additional support	9.1% (2)	
Mainstream with additional support	36.4% (8)	
Special unit	22.7% (5)	
Work type	**(n=7)**	
Higher education	4.5% (1)	
Full time employment	4.5% (1)	
Part time employment	9.1% (2)	
Supported employment	4.5% (1)	
Day options	4.5% (1)	
Other	4.5% (1)	
CP distribution		
R unilateral	36.4% (8)	
L unilateral	9.1% (2)	
Bilateral - lower limbs	31.8% (7)	
Bilateral - all four limbs	22.7% (5)	
Predominant motor type		
Spasticity	54.5% (12)	
Mixed	27.3% (6)	
Dyskinesia	18.2% (4)	

^cognitive impairment was determined by asking the young person/caregiver if a formal cognitive assessment had been undertaken. If yes, the result was recorded. If no, the caregiver was asked to suggest a level of cognitive impairment for the young person (none, mild, moderate, severe, unsure)

Table 6-5 Qualitative coding schema with definitions

Category	Subcategory	Subcodes	Description	n= (/22)
Comprehensibility			The participant's ability to understand the questions and response options	21
	Intended interpretation		The participant interpreted the item, response options, instructions, symbols as intended by the research team	20
	Unintended interpretation		The participant did not interpret the item, response options, instructions or symbols as intended by the research team, or it was unclear if it was interpreted correctly	10
		Accuracy of response	Interpreted accuracy of the participant's response based on observations by the interviewer, caregiver, and data analyser	4
		Additional clarification	Items or concepts which required additional explanation to be understood	7
		Observed challenges with comprehension	Observed difficulty understanding the item, tool or instructions based on observations by the interviewer, caregiver, and data analyser	10
		Literal interpretation	Items interpreted literally rather than more generally	2
	Visual presentation preferences		Specific preferences identified for visual presentation or changes to the visual presentation to enable understanding. This includes formatting and visual symbols	11

Category	Subcategory	Subcodes	Description	n= (/22)
	Response option preferences		Specific preferences identified for the response options	6
Feasibility			The participant's ability and willingness to complete the tool	12
	Missing items		Items not answered or unable to be answered by the participant. For example, considers a certain level of discomfort as normal and therefore does not associate pain with that activity	3
	Unreliable answers due to behaviour or distractibility		Answers to items seen as unreliable due to poor engagement, interpreted as due to behaviour, fatigue or distractibility	4
	Version preference		Preference expressed or observed for *Talking Mat* or Pen/paper version	12

Comprehensibility

Data relating to comprehensibility were coded from twenty-one children and young people. Comprehensibility data were further coded to four sub-categories, which included intended or unintended interpretation of the tools. An item matrix table tracking all unintended interpretations of items, response options and instructions, along with the interpretation of the research team and advisory group and any changes made can be found in Appendix E3. No interpretation issues identified were solely related to a particular administration method (*TalkingMat* vs paper). All the interpretation issues identified were relevant to both, as they related to the item wording, item examples, response option wording or visual symbols. Consensus EFFC scores were >21 for n= 17/22 (mBPI) and n= 15/19 (FOPQ-C-SF), indicating that most children and young people appropriately communicated responses to the tools. The qualitative data also showed that most items and response options were interpreted as intended (n=20), as shown by one person's reasoning for selecting 2 = 'a bit' (Figure 6-1):

"Just very angry sometimes when my leg starts to hurt.... just sometimes but not all the time, so I just chose the middle one because half the time it does and half the time it doesn't."

18 yo male, CFCS III, moderate CI, mBPI paper

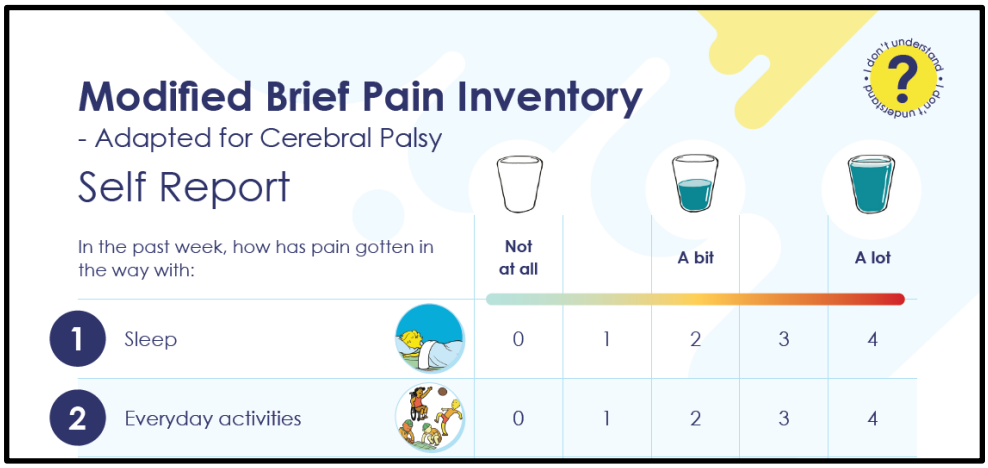


Figure 6-1 Example of the first two items and response scale of the mBPI

Symbols used with permission ©Talking Mats® 2023.
***Abbreviations*: *mBPI* =** *modified Brief Pain Inventory*

The inclusion of standardised examples for items that were more general, along with the visual symbols, appeared to improve accuracy of interpretation. Children and young people demonstrated understanding of even abstract items, for example when answering 'pain makes me think something bad will happen' (FOPQ-C-SF item 9 –Figure 6-2):



Figure 6-2 FOPQ-C-SF item 9 image ("Pain makes me think something bad will happen"

Used with permission, ©Talking Mats® 2023.
Abbreviations*: *FOPQ-C-SF *= Fear of Pain Questionnaire for Children Short Form*

Possible reasons for unintended interpretation

Within the unintended interpretation sub-category, three sub-codes were identified as possible contributors: accuracy of response, observed challenges with comprehension and additional clarification. The five children and young people who did not reach the effectiveness threshold on the EFFC all had at least one item coded as 'unintended interpretation', demonstrating both reduced understanding and engagement. It was difficult to be certain of the response accuracy in cases where cognitive ability was low or behaviour prohibited engagement, for example when a young person was trying to complete the questions quickly. For one young person, it was not clear if they were making a joke or if they did not understand the item 'how much has pain gotten in the way of school' (Appendix E5, quote 3 – mBPI Talking Mat). For children and young people who used AAC, parents provided suggestions to clarify the accuracy of the response (i.e. wording or symbol explanation). In relation to selecting a response option symbol for the mBPI:

"I think a water glass [symbol] would work. [She] will typically look and then look away for a bit. She'll look like she's not engaged but what she's doing is processing. Usually, it just takes a little while."

Parent of 20 yo female, CFCS III (AAC user), moderate CI, mBPI Talking Mat

Additional clarification by providing further examples was suggested for two mBPI items. Two children and young people suggested alternative examples for everyday activities (mBPI item 2), including caring for pets (Appendix E5, quotes 1 and 11). Picture symbols were generally well received, however four children and young people, all with cognitive impairment, initially interpreted the visual symbols literally by focusing on the task itself rather than how pain interferes with it. When looking at the symbol for 'looking after myself' (picture of a person brushing their teeth):

"Facilitator: What about pain getting in the way of you helping to look after yourself?

Participant: I brush my teeth.

Facilitator: You brush your teeth.

Participant: I was brushing my teeth in the bathroom."

11 yo, CFCS II, moderate CI, mBPI Talking Mat

Response option preferences & visual presentation preferences

Children and young people were explicitly asked to identify preferences between circles or a water glass for the mBPI response option symbols (Appendix E2). All except one preferred the water glasses or did not have a preference. An empty glass symbol was recommended for 'not at all' rather than the original image of a glass with a small amount of water in it (Figure 6-1).

Feasibility

Completion times and missing items

Median completion times were 6.2 min (IQR=5.3-7.6) for mBPI and 4.1 min (IQR=2.7-4.9) for FOPQ. Completion time for AAC users and those with cognitive impairment increased by +4.2 min (mBPI), +9.6 min (FOPQ) and +1.8min (mBPI), +1.4min (FOPQ), respectively. Completion of *TalkingMats* versions took longer than paper (+2.3mins mBPI, +1.1mins FOPQ). Table 6-6 and Figure 6-3 show the median completion times for each tool, by category of participant. Figure 6-3 visualises the increased time taken for AAC users, with one participant taking up to 24 minutes to complete the mBPI. The extended time resulted from the participant needing additional processing time for each item and requiring breaks during the interview due to fatigue. Subsequently, they did not fully complete the FOPQ-C-SF. A further two children and young people (one AAC user and one verbal communicator) did not fully complete the FOPQC-S-F due to challenging behaviour and parent recommendation. Although all children and young people completed the mBPI, seven missing items (9.8% of total data) were identified in three assessments: everyday activities (n=1), school/work/day activities (n=1), looking after myself (n=2), having fun (n=1), mobility (n=1) and favourite thing (n=1). Of the participants with missing items, all had reduced communication ability with unfamiliar people (CFCS level III and IV) and cognitive impairment. Identified reasons for missing items included difficulty disentangling the impact of pain and impact of disability (mBPI – mobility and looking after myself), inability to choose one favourite thing (mBPI - favourite thing), difficulty understanding abstract questions due to cognitive limitations

(mBPI - having fun) and lack of relevance as the individual did not attend school, work or respite due to complex health needs (mBPI - school/work). For example, after a young person did not answer 'looking after myself' (mBPI item 6), the caregiver speculated this may be because it is difficult to separate the impact of disability and impact of pain as the young person has significant motor limitations:

"Whereas I would say, from my perspective here, pain does affect her capacity to let us do things or participate in her own care. She really doesn't want self-care to happen when she's sore... she's like 'I want to stay in bed, leave me alone'"

Parent of 20 yo female, CFCS III, moderate CI, mBPI Talking Mat

Another young person expressed difficulty knowing if they were answering the questions correctly, because they were 'so used to being in pain':

I'm so used to being in pain but it also depends on how much you've pushed yourself, how much physical activity you've done. I do have actually [have] a bit of a [high] pain tolerance, due to all the other stuff I've been through. So I may not be able to give the best answers.

14 yo female, CFCS I, no CI, mBPI paper

For one younger child, 'pain makes me not want to make plans' (FOPQ item 7) was not felt to be relevant, however this was still answered by all children and young people.

Facilitator: Pain makes me not want to make plans.

Participant: No. Not me.

Facilitator: You still like to make plans?

Participant: I don't really make plans [Mum makes them]

6 yo female, CFCS I, no CI, FOPQ-C paper

Table 6-6 Median duration completion times by tool and category

Category	mBPI median (IQR)	FOPQ median (IQR)
Overall	6.2 (5.3-7.6)	4.1 (2.7-4.9)
Verbal	6.2 (4.8-7.4)	3.7 (2.5-4.6)
AAC user	10.4 (9.6-11.2)	13.3 (9.7-16.9)
CI	7.6 (6.1-9.1)	4.5 (4.2-5.2)
No CI	5.8 (4.2-7)	3.1 (1.8-4.8)
Pen/paper	5.8 (4.2-6.5)	3.5 (2.3-3.9)
Talking Mat	8.1 (6.1-9.1)	4.6 (3.1-5)

***Abbreviations : AAC** = augmentative and alternative communication, **CI** = cognitive impairment, **FOPQ** = Fear of Pain Questionnaire for Children Short Form adapted for cerebral palsy, **MBPI** = modified brief pain inventory adapted for cerebral palsy*

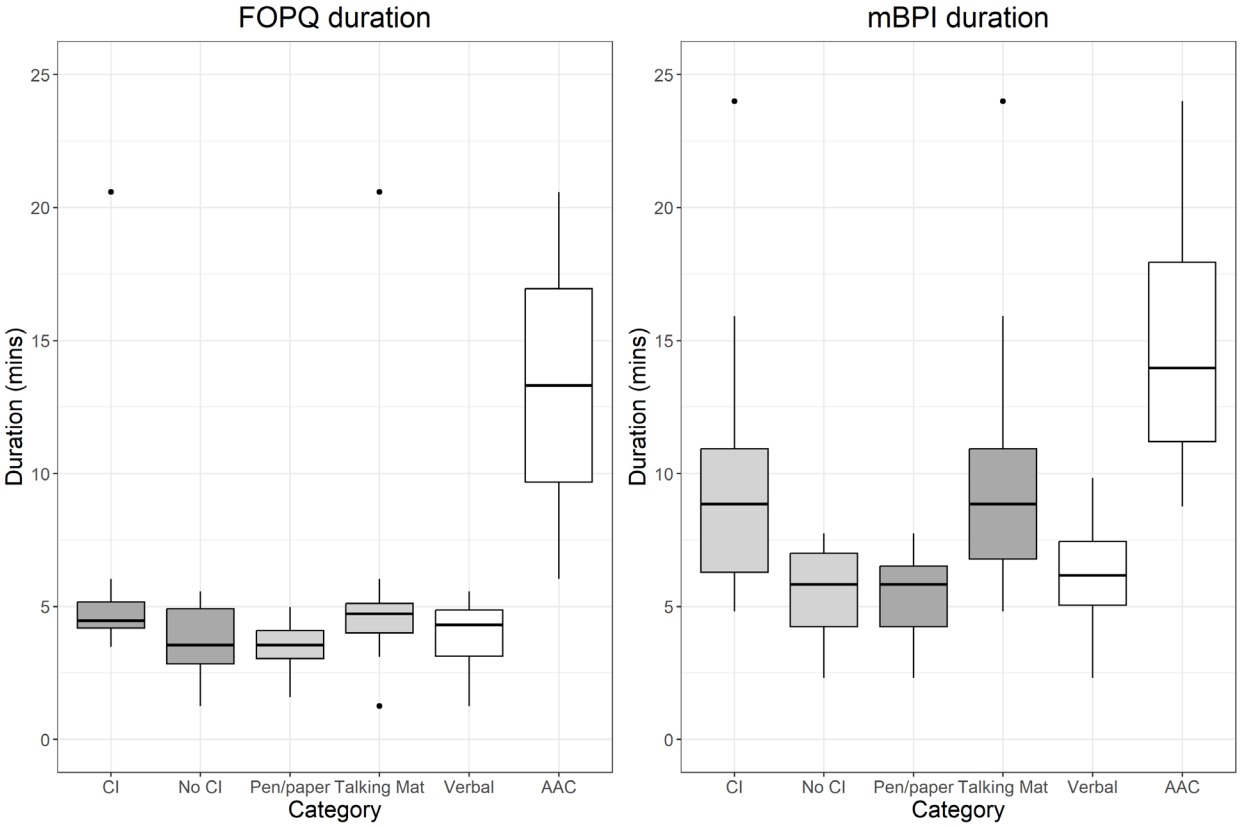


Figure 6-3 Boxplot of completion times of the adapted Fear of Pain Questionnaire Short Form (FOPQ) and Modified Brief Pain Inventory (mBPI), comparing differing communication needs, cognitive abilities and versions of the tools.

***Abbreviations: AAC** = augmentative and alternative communication, **CI** = cognitive impairment, **FOPQ** = Fear of Pain Questionnaire for Children Short Form adapted for cerebral palsy, **MBPI** = modified brief pain inventory adapted for cerebral palsy*

Version preference

Accessing picture symbols was challenging for one young person who also had a visual impairment, reporting he 'just listened' to the questions as he could not see the pictures:

Facilitator: The other thing that I wanted to know is what you thought of the pictures. Because I don't know if you were using them, or whether you were just listening.

Parent: More to say, categories. Actions. Sensory actions... We're wondering, all the pictures that were on the cards, whether you could see them, whether you were looking at the pictures, or more to say, go to categories. Turn the page. Something's wrong. Turn the page. You couldn't see them. Listening, could not see

Participant: [Nods, indicating yes]

Parent: So, with the pictures... you couldn't see them?

Participant: [Nods, indicating yes].

Facilitator: You couldn't see them... So, you were listening?

Parent: Categories, action words. Sensory actions. Were you listening to the descriptions? Is that how you were answering the questions, using your listening?

Participant: [Nods, indicating yes].

11 yo male, CFCS III (AAC user), no CI, mBPI/FOPQ Talking Mat

The paper version was preferred for older children and those without cognitive impairment, however all were still able engage with the TalkingMats version. TalkingMats (Figure 6-4) was preferred by younger children, AAC users and those with cognitive impairment (quote 24, Appendix E3).



Figure 6-4 Example of a completed Talking Mat for the mBPI.

Symbols used with permission. ©Talking Mats® 2023.
***Abbreviations*: *mBPI* =** *modified Brief Pain Inventory*

Further suggested changes

Twenty-two suggested changes were identified during inductive coding (Appendix E6). Implemented changes are listed in Table 6-7, with item progression from original to final tracked in Appendix E3.

Table 6-7 Changes implemented after data review by author team and consumer advisory group

Implemented changes	Related category	Tool	Version
Remove 'day activities' from school/work/day activities as this caused confusion with mBPI item 2 'everyday activities'. Instead include 'day options/respite' as an example in the administration guide	Comprehensibility	mBPI	Both *Talking Mat* and paper

Implemented changes	Related category	Tool	Version
Alternate background row colour	Comprehensibility	mBPI & FOPQ-C	Paper
Make font slightly larger	Feasibility	mBPI & FOPQ-C	Paper
Put a line either side of the middle response option on the paper version	Comprehensibility	mBPI & FOPQ-C	Paper
Simplify introduction for the FOPQ-C-SF	Comprehensibility	FOPQ-C	*Talking Mat* & paper
Use an empty glass image rather than slightly full for the 'not at all' response option for the mBPI	Comprehensibility	mBPI	*Talking Mat* & paper
Use the water glass images over the circle images for the mBPI	Comprehensibility	mBPI	*Talking Mat* & paper
Have FOPQ *Talking Mat* response options on the same side as the paper version (i.e. 'this is not me on the left' and 'this is me on the right' – **Figure 6-5**)	Comprehensibility	FOPQ-C	*Talking Mat*
Initially remove the middle symbol from the *Talking Mat* if an individual struggles to choose from three options due to cognitive ability	Feasibility	mBPI, FOPQ-C	*Talking Mat*
Add 'or helping to look after myself' to mBPI item 6 (looking after myself) for those who have limited independence	Comprehensibility	mBPI	*Talking Mat* & paper

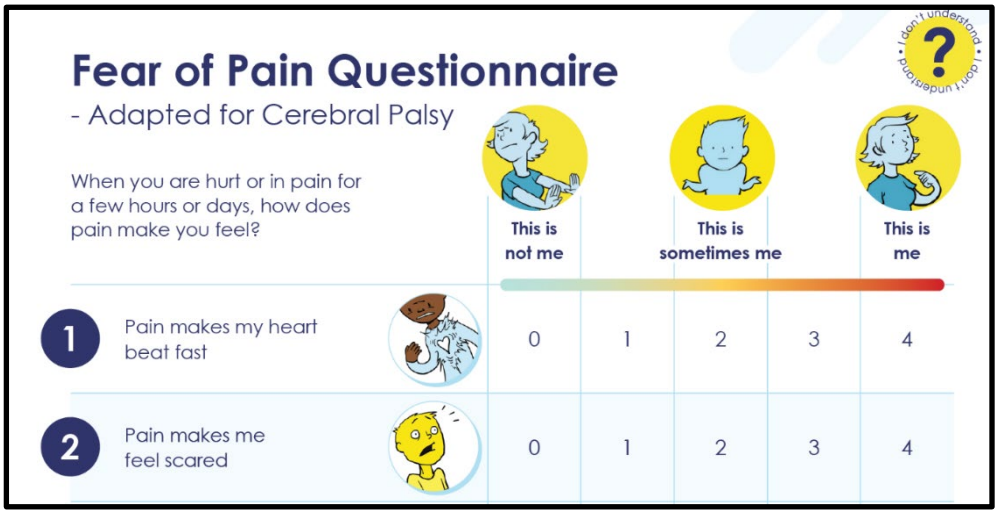


Figure 6-5 Example of the first two items and response scale of the FOPQ-C-SF.

Symbols used with permission ©Talking Mats® 2023.
Abbreviations*: *FOPQ-C-SF *= Fear of Pain Questionnaire for Children Short Form*

6.7 Discussion

This study examined the comprehensibility and feasibility of adapted versions of the mBPI and FOPQ-C-SF in children and young people with CP and varying motor, communication and cognitive abilities. The study found the adapted tools are both comprehensible and feasible for most children and young people with CP, including AAC users and those with mild-moderate cognitive impairment. Ten minor adjustments will be implemented prior to wider testing, and as missing items made up less than 10% of the data, no items will be deleted [31]. From a clinical perspective, this means that children and young people with CP can and should be encouraged to provide self-reported pain information using these newly adapted tools. Regular use of these adapted tools in clinical practice may provide the first step in identifying self-reported pain interference and pain-related fear in children and young people with CP, subsequently facilitating access to best-practice chronic pain interventions, such as positive pain coping strategies, which can improve quality of life [26, 34].

When assessing comprehensibility, approximately three quarters of children and young people demonstrated 'effective communication' on the EFFC while completing the mBPI and FOPQ-C-SF. The ability to engage with the assessment was also dependent on how concrete or abstract the concepts were. The FOPQ-C-SF had lower EFFC scores

and fewer full completions than the mBPI, likely because almost all the items are abstract in nature. Literal interpretation of the item symbols was a common issue for young people with a cognitive impairment in this study, and was similarly noted in a previous study adapting a PROM for people with intellectual disability. [253] This was managed by providing additional examples to help the participant relate the symbol to the intended activity. For the participants in our study who did not meet the 'effectiveness' threshold, the assessment still provided a degree of valuable self-reported information that was otherwise unknown. This demonstrates the importance of exploring a young person's own pain perspectives as a right, regardless of age or cognitive ability [209, 248]. It is therefore recommended that self-report tools are used as first priority, and if the young person is unable to complete the whole tool or there is concern about self-report accuracy, a caregiver can provide an additional proxy report. This ensures proxy-report is not the default, as parents may find it difficult to estimate the impact of their child's pain [261]. In this study, the interplay between behaviour, fatigue, concept abstractness and familiarity with the interviewer likely impacted EFFC scores, subsequent 'effective communication' rating and overall comprehensibility. In clinical practice, strategies can be implemented to improve 'effective communication' when using the tools, such as being in a familiar environment with someone known to the young person.

Two of the three people with missing items were communicating using AAC and were unable to provide reasons for not responding to these items. In this case, caregivers suggested reasons for the missing items, which included difficulty disentangling the impact of pain from the other impacts of disability. For people with significant functional limitations, cognitive impairment and/or other medical conditions, the challenge of isolating the impact of pain was similar to findings by Rinde et al. 2023 [261], who explored the theme 'my child's pain is just one piece of a complex jigsaw puzzle'. Some people in the present study also had difficulty identifying the impact of pain because pain is a usual part of life, consistent with other literature investigating pain in children and CP [261]. Caregivers were helpful in these circumstances as they were able to suggest alternative phrases the young person might typically use instead of pain, such as 'think about when your leg is stiff'.

The feasibility of completing the assessments was highly dependent on the administration method used. While it was originally planned to ask all participants to complete both administration methods of each tool (i.e. *TalkingMat* and paper version of the mBPI, and *TalkingMat* and paper version of the FOPQ-C), the advisory group recommended against this due to participant burden. Where possible, all participants completed one tool in the *TalkingMat* version and the other in the paper version to allow comparison between the administration methods (i.e. mBPI *TalkingMat* and FOPQ-C paper). The paper versions of the tools can be used with the support of a freely available administration guide, however *TalkingMats Ltd* requires practitioners to complete training prior to use in clinical practice. The training is low cost, accessible online and self-paced. As a result, it is unlikely to pose a significant barrier to implementation in clinical settings. The training also has broader applicability for clinicians working with people with communication disability and for people with cognitive impairment (with or without speech). *TalkingMats* was the preferred version for AAC users; however, it is still important to be flexible for individual needs. For one young person, a suggestion was made to use high-contrast symbols like those in their Pragmatic Organisation of Dynamic Display (PODD) to meet their sensory requirement. This need for flexibility was identified by parents of AAC users in our previous study [211] and is unsurprising given the high level of customisation required when developing and prescribing AAC systems [262, 263].Completion times increased for children and young people with cognitive impairment and communication access needs, with the mBPI taking up to 24 mins and FOPQ up to 21 mins for one AAC user. A key accessibility feature for those who use AAC is allowing them adequate time to engage [259]. Clinicians should consider how the young person can best self-report when completing the assessment and employ strategies such as completing the tool during the appointment with a clinician. This will also enable richer discussion around pain experience and management, goal setting and shared decision making. Furthermore, providing the assessment questions ahead of time can enhance communication rate for AAC users with CP, while also addressing cognitive load and associated fatigue issues [259]. The motor impairments in CP increase the complexity around operational competencies for using the AAC system, making the rate of communication slower and more fatiguing. Clinicians should reflect on their individual clinical setting and prioritise self-reporting of pain, particularly for those who need more time to respond. In time-limited clinical settings, one option is for the young

person to complete the assessment with a family member and send it ahead of time to the healthcare team. However, it is essential to allocate time during the appointment to review the completed assessment and discuss its implications. It may also be useful to complete the adapted tools within broader assessments, such as surveillance services like CP-UP in Sweden.[264]

Adapting pain assessments for those who use AAC, younger age groups and those with cognitive impairment did not change their usability or comprehensibility more broadly. This is consistent with the concept of universal design, demonstrating how improved accessibility for those with specific needs also improves usability for those without [248]. Children and young people without cognitive or communication impairment reported that the simplified language and response scales, improved formatting and images made the tools quicker and easier to use. While the adaptations have enabled at least a degree of self-report for those with mild-moderate cognitive impairment, we did not have any children and young people with severe cognitive impairment in the study, even though they were not excluded. Our observations during the study suggest it is unlikely a young person with severe cognitive impairment would have the capacity to complete the adapted assessments, although this needs to be determined. Observational tools, such as the Paediatric Pain Profile [265] may be most useful in these cases. A strength of this study was including AAC users, younger children and those with cognitive impairment, groups which are often excluded in research due to a perception that they are unable to self-report [266]. There is a natural flow-on effect from exclusion in research to exclusion in clinical practice, with most regularly used PROMs requiring verbal or written communication, and cognitive abilities of >8 years of age [247]. Our study showed the adapted pain assessment tools are suitable for individuals as young as five years, those with mild-moderate cognitive impairment, and diverse motor and communication abilities. The broader age range also allowed comparison between general preferences for administration method, with younger children generally preferring the *TalkingMats* version and older participants generally preferring pen/paper.

While previous studies have adapted or investigated PROMs for people with intellectual disability[253, 267], we are not aware of any studies that have considered adaptations for a heterogenous group like CP, which includes people with diverse cognitive, communication and motor abilities. Participant diversity was important in identifying

further refinements to the tools (Table 6-7). For example, the inclusion of 'helping to look after myself' to mBPI item 6 improves inclusivity for people with limited independence. Furthermore, strategies to reduce visual demand - such as alternating the background row colour and increasing the font size - were identified by participants with visual impairments. Similar to our study, other research has successfully improved comprehensibility by simplifying response options and adding visual symbols for younger children and people with intellectual disability [251, 253]. A unique feature of our adaptation process was the use of *TalkingMats* as a PROM administration method rather than solely a communication tool. Other research with children with intellectual disability has used *TalkingMats* alongside other arts based participatory methods, such as body mapping and photo voice, to identify key constructs of interest for patient-reported experience measures [267].

Although the study participants were skewed towards higher levels of functioning (i.e. GMFCS/MACS levels I-III), there were still sufficient participants in the lower functioning groups to be confident that saturation was reached. COSMIN consider a sample size of 4-6 participants as adequate [240], and the study included four participants in GMFCS levels IV/V and 6 participants in MACS levels IV/V. Furthermore, even from a highly heterogenous group, there were few changes to be made to the items, wording, response options and administration (Online Resource 3). Given there were so few changes to be made from such a diverse group and considering the sample size both overall and for each subgroup of participants meets the COSMIN recommendations for 'very good' or 'adequate', we are confident the tool is appropriate for use across the diversity of ability.

The study had several limitations. It is usual for multiple smaller rounds of pilot testing to be completed with changes made to the tool between rounds [31]. Given the heterogeneity of the CP population, we felt it more appropriate to initially capture the perspectives of a larger group, rather than making changes from a smaller group and potentially needing to undo those changes [211]. We had planned to complete additional pilot testing if wording changes were identified [255]. However, the only change implemented was the removal/addition of one word, and as such it was not deemed necessary to re-test.

It is possible that researcher bias influenced the analysis of observed body language/non-verbal cues, however this was minimised by involving at least two researchers at each analysis stage, all with experience working with people with CP and an experienced speech pathologist to specifically analyse AAC users. A further limitation was potential bias from having a communication support person and/or parents present during the interviews. To minimise this, interviewers spoke with the support person prior to the interview about focusing on the young person's perspective and voice. For AAC users, the support person can also be considered a benefit given their greater familiarity with the person's communication cues. A final limitation was possible fatigue experienced by the children and young people after completing two tools in one session. This may have impacted engagement with the second tool, however this was managed by allowing breaks.

Future directions

The mBPI and FOPQ-C-SF adapted for children and young people with CP will now undergo further psychometric testing prior to being made freely available for use in clinical practice and research. It is possible these tools could be digitised in future, potentially decreasing administration times and allowing greater customisation. It is important to consider how a digitised version could be used alongside an AAC device, and how it would be sustainably maintained and freely accessed.

6.8 Conclusion

The adapted mBPI and FOPQ-C-SF are likely comprehensible and feasible for use in children and young people with CP, including those with diverse cognitive and communication abilities. Tool adaptations such as alternative administration methods, visual symbols and wording simplification helped improve self-report participation for people with CP. Minor changes have been identified through the pilot testing process and will be implemented prior to further psychometric testing. Prioritising the inclusion of people with diverse cognitive and communication abilities is both feasible and critical in developing inclusive and accessible assessment tools.

Chapter 7 – The Pain Interference Questionnaire and Fear of Pain Questionnaire for children and young people with cerebral palsy: examination of structural validity (Study 5)

7.1 Preamble

Chapter 6 reported the results of the pilot testing two new tools based on the FOPQ-C-SF and mBPI for young people with cerebral palsy (CP). Utilising modified cognitive interviews, this study showed the CP-adapted versions are comprehensible and feasible for young people with CP who also have mild-moderate cognitive impairment or complex communication needs. The combined findings of Chapters 4-6 provide sufficient evidence of content validity for these tools in the CP population, including relevance, comprehensiveness and comprehensibility. The FOPQ-C-SF and mBPI were subsequently renamed as the Fear of Pain Questionnaire for Cerebral Palsy (FOPQ-CP) and the Pain Interference Questionnaire for Cerebral Palsy (PIQ-CP), respectively.

In order to recommend an assessment tool for use, the newly updated COSMIN guidelines (2024) advise that, ideally, all psychometric properties should be investigated and found to have sufficient evidence.[222] However, work by Smith N and colleagues shows clearly that no tools assessing the impact of pain on emotional functioning are close to having sufficient evidence for all psychometric properties, given limited research in this area.[113] In the case of no tool having sufficient evidence for all psychometric properties, it is possible to recommend a tool for use based on the content validity quality.[222] The previous COSMIN guidelines (2018), on which this thesis was originally based, advised the second-most important psychometric property to investigate was internal consistency.[27] For this reason, a cross-sectional study was designed to determine the structural validity and internal consistency of the FOPQ-CP and PIQ-CP for young people with CP. Chapter 7 provides the results of a cross-sectional study aiming to determine the structural validity of the FOPQ-CP and PIQ-CP. The final versions of the tools, along with the administration guides, are provided in Appendix A: CP-adapted pain assessment tools.

7.2 Authorship declaration

This paper has been submitted for publication.

Smith, M.G.[1,2,3], Gibson, R.J.[1], Russo, R.N.[4,5], Smith, N.[6,7], Harvey, A.R[8,9]. The Pain Interference Questionnaire and Fear of Pain Questionnaire for children and young people with cerebral palsy: examination of structural validity. *Submitted for publication*

[1]School of Allied Health Science and Practice, University of Adelaide, Adelaide, Australia;

[2]Novita, Adelaide, Australia;

[3]Women's and Children's Hospital, Adelaide, Australia;

[4]Paediatric Rehabilitation Department, Women's and Children's Hospital, Adelaide, Australia;

[5]College of Medicine and Public Health, Flinders University, Adelaide, Australia;

[6]Kids Rehab Perth Children's Hospital, Nedlands, Australia

[7]The University of Western Australia, Crawley, Australia

[8]Neurodisability and Rehabilitation, Murdoch Children's Research Institute, Melbourne, Australia;

[9]Medicine, Dentistry and Health Sciences, University of Melbourne, Melbourne, Australia

Statement of Authorship

Title of Paper	The Pain Interference Questionnaire and Fear of Pain Questionnaire for children and young people with cerebral palsy: examination of structural validity
Publication Status	☐ Published ☐ Accepted for Publication X Submitted for Publication ☐ Unpublished and Unsubmitted work written in manuscript style
Publication Details	Smith, M.G.[1,2,3], Gibson, R.J.[1], Russo, R.N.[4,5], Smith, N.[6,7], Harvey, A.R[8,9]. The Pain Interference Questionnaire and Fear of Pain Questionnaire for children and young people with cerebral palsy: examination of structural validity. *Submitted for publication*

Principal Author

Name of Principal Author (Candidate)	Meredith Smith		
Contribution to the Paper	Completed data collection, analysis of all data, wrote manuscript and acted as corresponding author		
Overall percentage (%)	65%		
Certification:	This paper reports on original research I conducted during the period of my Higher Degree by Research candidature and is not subject to any obligations or contractual agreements with a third party that would constrain its inclusion in this thesis. I am the primary author of this paper.		
Signature		Date	6/1/25

Co-Author Contributions

By signing the Statement of Authorship, each author certifies that:

i. the candidate's stated contribution to the publication is accurate (as detailed above);

ii. permission is granted for the candidate in include the publication in the thesis; and

iii. the sum of all co-author contributions is equal to 100% less the candidate's stated contribution.

Name of Co-Author	5% Nadine Smith		
Contribution to the Paper	Contributed to data collection, edited manuscript		
Signature		Date	14/1/25
Name of Co-Author	10% Adrienne Harvey		
Contribution to the Paper	Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	13-01-2025
Name of Co-Author	10% Rachel Gibson		
Contribution to the Paper	Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	10/2/25
Name of Co-Author	10% Ray Russo		
Contribution to the Paper	Contributed to data analysis, supervised development of work, edited manuscript		
Signature		Date	6/1/2025

7.3 Abstract

Purpose: Few valid and accessible chronic pain assessment tools exist for people with cerebral palsy (CP). Two tools have recently demonstrated content validity for children and young people with CP: The Pain Interference Questionnaire for Cerebral Palsy (PIQ-CP) and the Fear of Pain Questionnaire for Cerebral Palsy (FOPQ-CP). This study aimed to evaluate their structural validity and describe how many participants could self-report using the adapted tools, including those with diverse cognitive and communication abilities.

Materials and Methods: A prospective cross-sectional multi-site study was undertaken. People with CP aged 5-30 years completed the PIQ-CP and FOPQ-CP. Confirmatory Factor Analysis (CFA) assessed structural validity. Data were analysed to report the proportion able to self-report.

Results: Data from 128 participants fit the hypothesised one-factor (PIQ-CP) and two-factor (FOPQ-CP) structure (CFI>0.96 or SRMR<0.09). Most (79%) could self-report, 11% required additional parent-report because the self-report accuracy was uncertain and 10% required parent-report only, due to severe cognitive impairment.

Conclusions: The PIQ-CP and FOPQ-CP have sufficient structural validity and provide access to self-report in most children and young people with CP. They can now be used to support equitable assessment of the impact of chronic pain in young people with CP, including those with diverse abilities.

7.4 Introduction

Accurate assessment of chronic pain within a biopsychosocial framework is an important first step in identifying appropriate treatment options for children and young people with chronic pain.[20] While tools to comprehensively assess chronic pain are freely available and well-researched in the general paediatric population[97, 98], there are few valid, appropriate and accessible tools for children and young people with disabilities, particularly those with diverse communication, cognitive and motor abilities.[23, 25, 112, 210] To address this gap, two existing pain assessment tools, the modified Brief Pain Inventory[154] and the Fear of Pain Questionnaire for Children Short Form (FOPQ-C-SF)[235] have been recently adapted by our research group[211, 268] to be more appropriate and accessible for children and young people with cerebral palsy (CP), the most common cause of childhood physical disability globally[1]. Several of the modifications made aim to improve access to self-report for children and young people with CP and with diverse cognitive and communication abilities. The newly adapted tools are named the Pain Interference Questionnaire for Cerebral Palsy (PIQ-CP) (referred to in a previous publication as the modified Brief Pain Inventory adapted for Cerebral Palsy), and the Fear of Pain Questionnaire for Cerebral Palsy (FOPQ-CP), which assess the constructs of pain interference and pain-related fear/activity avoidance, respectively. Both tools are patient-reported outcome measures (PROMs), which means they rely on self-report from the individual. They also both cover several of the recommended pain domains for children and young people with CP, including impact of pain on emotional functioning, physical function, participation, sleep, and quality of life.[203] Assessment of these domains is key in identifying children and young people who may benefit from best practice multidisciplinary interventions for chronic pain, which are currently underutilised in CP.[34, 100]

The PIQ-CP and FOPQ-CP have undergone rigorous content validity evaluation to ensure suitability for children and young people with CP aged 5-30 years.[211, 269, 270] After establishing adequate content validity, two of the priority psychometric properties for PROMs are structural validity and internal consistency.[244] Both the PIQ-CP and FOPQ-CP are PROMs based on a reflective measurement model, where all items are intended to represent expressions of the same underlying construct.[244] Consequently, internal consistency and structural validity are inter-related. Structural validity is a psychometric property within the broader 'validity' domain, and measures the extent to

which scores on a PROM appropriately represent the dimensionality of the construct being measured.[271] Internal consistency is a property within the broader 'reliability' domain and measures how well the items within a scale or subscale relate to each other.[271]

The primary aim of this study was to determine the structural validity of the PIQ-CP and FOPQ-CP for children and young people with CP. We hypothesised the PIQ-CP would support a one-factor structure[154], and the FOPQ-CP a two-factor structure[235], given the confirmatory factor analysis (CFA) results of the original versions. The secondary aim of this study was to describe the extent to which self-report for children and young people with CP and with diverse cognitive and communication abilities was possible using the PIQ-CP and FOPQ-CP, measured by how many were able to complete the self-report versions of the tools.

7.5 Materials and Methods

Study design

A prospective cross-sectional multi-site study design was used. This study received approval from the Women's and Children's Human Research Ethics Committee (2022/HRE10554). The study was guided by a lived experience advisory group, including two young adults with CP and a parent of a child with CP. The advisory group met bimonthly throughout the project and were reimbursed for their time.

Participants and recruitment

Recruitment took place at three Australian sites: Novita Disability Services (South Australia), Women's and Children's Hospital Paediatric Rehabilitation Department (South Australia) and Perth Children's Hospital Kids Rehab (Western Australia) from November 2023 – November 2024. Eligible children and young people aged 5-30 years of age with CP (Table 7-1) were identified by clinicians at each site through screening outpatient clinic lists and electronic medical record. Both tools have demonstrated content validity for this age group[270], and the advisory group specifically recommended extending the cut off for young adulthood to 30 years to account for those with disability who may experience delayed transitions to adulthood social roles. Written informed

consent was obtained from young adults without cognitive impairment, caregivers of children and caregivers of young adults with cognitive impairment.

Table 7-1 Inclusion and exclusion criteria

INCLUSION CRITERIA	EXCLUSION CRITERIA
Individuals with cerebral palsy aged 5-30 years of age	Children with cerebral palsy who are less than 5 years of age
Parents of children with cerebral palsy (5-30 years of age)	
To complete the self-report version, the individual must be able to: • Answer at least 10 questions about the impact of their pain on their life • Indicate responses using a reliable method for the individual. This could include using visual symbols, verbal, written or augmentative/alternative communication device options	
Parent report versions will only be completed where: • Both the parent/caregiver and the therapist are concerned about the accuracy of the individual's self-report version • For individuals who are unable to self-report pain by any method	

Sample size

The target sample size was at least 100 individuals completing the self-report version of the PIQ-CP and FOPQ-CP, which is considered 'very good' by COSMIN.[240]

Participant demographics

Demographic characteristics (age, sex, predominant motor type, communication ability, gross motor ability and cognitive ability) were provided by the child/young person or their caregiver. Communication and gross motor ability were classified using the Communication Function Classification System (CFCS)[16] and the Gross Motor Function Classification System (GMFCS)[14] respectively, two commonly used systems

for describing functional abilities in CP. Each classification provides a 5-level rating from I (least impaired) to V (most impaired). Children and young people were asked if they had ever experienced pain lasting longer than a few weeks.

Measures

Pain Interference Questionnaire for Cerebral Palsy

The PIQ-CP was used by children and young people to report pain interference. The PIQ-CP was available in two formats– standard pen/paper or *Talking Mat®*. *Talking Mats®* is an evidence based visual communication framework developed to improve the expression of views of people with disability.[229] Both formats asked for a response to the same 12 items using the lead in phrase 'in the past week, how has pain gotten in the way with….'. Responses were provided on a 5-point ordinal scale (0='not at all' to 4 = 'a lot'), with scores totalled to produce a sum score (0-48). The *Talking Mats®®* response scale contained three symbols, with each item able to be placed either under a symbol or in between the symbols, producing a 5-point scale consistent with the paper version. Higher scores indicated greater pain interference. A parent-report version of the PIQ-CP was also available in the same format.

Fear of Pain Questionnaire for Cerebral Palsy

The adapted version of the FOPQ-CP was used by children and young people to report pain-related fear and activity avoidance. The FOPQ-CP was also available in two formats – standard pen/paper or *Talking Mat®*. Both versions asked for a response to 10 statements about how pain makes you feel, with responses provided on a 5-point ordinal scale (0='this is not me' to 4 = 'this is me'). Scores were totalled to produce a sum score (0-40), with higher scores indicating greater pain-related fear and activity avoidance. There is no parent report version of the FOPQ-CP, as a previous study found the construct of pain-related fear is too personal to be answered accurately by a proxy.[211] A previous CFA of the original FOPQ-C-SF yielded a correlated two-factor structure, including a 'fear of pain' factor and 'avoidance of activities' factor (Appendix F1).[235] The correlated structure was consistent with the known links between pain-related fear and activity avoidance seen in the fear-avoidance model of chronic pain.[272, 273]

Procedure

Data was collected with a clinician, either as part of usual clinical practice, or a stand-alone research appointment. Administration method (*Talking Mat*® or pen/paper) was selected based on the decision-making tree (Figure 7-1). Self-report was prioritised wherever possible, with parent-report used only if the person was unable to self-report at all, or the clinician and caregiver were uncertain about the accuracy of the person's self-report (Table 7-1). Support was provided as needed by the clinician and/or caregiver to allow the child or young person to self-report, such as reading questions out loud or providing assistance to physically place/circle response options. All clinicians involved in data collection were provided with training by the author (MS). Completed assessments were uploaded to REDCap electronic data capture tool hosted at the University of Adelaide.[215, 216]

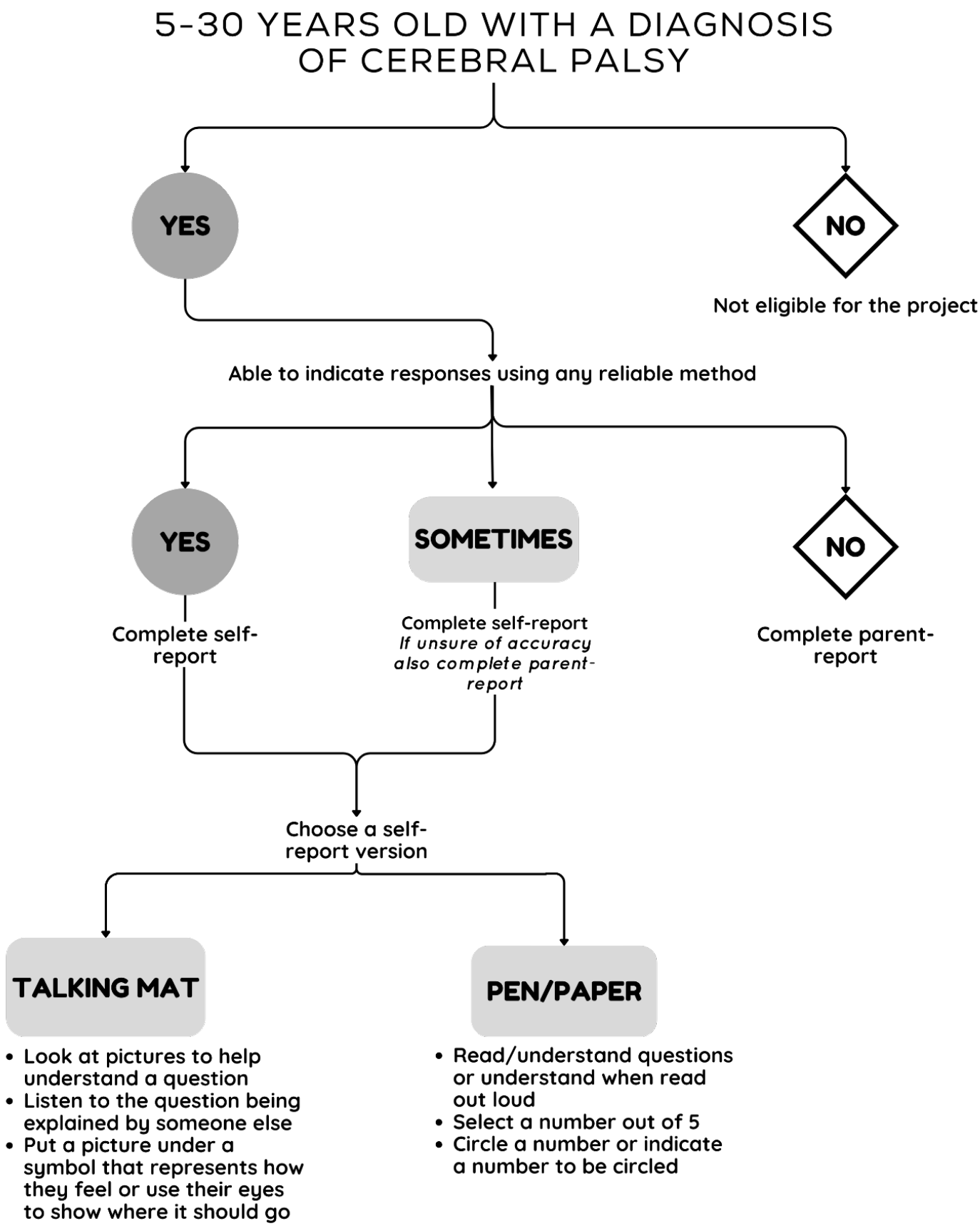


Figure 7-1 Decision making tree to determine the most appropriate version of the tool to complete

Data analysis

Statistical analysis was completed using R software.[274] Summary descriptive statistics were used to describe the characteristics of the dataset. Item level analysis was conducted to identify items with extensive missing data. The data on missing responses were examined for each item in the two assessments, with items with >15% missing data identified for further investigation.[31, 275]

Confirmatory factor analysis

The underlying factor structure of the PIQ-CP and FOPQ-CP was examined using confirmatory factor analysis (CFA). CFA was conducted on the PIQ-CP and the FOPQ-CP using self-report data only. A two-factor measurement model, allowing for correlations, was used for the FOPQ-CP, and a one-factor measurement model for the PIQ-CP, based on previous CFAs of the original tools.[154, 235] The model was fitted using the Lavaan package[276] with the Weighted Least Squares with Mean and Variance adjustment (WLSMV) estimation, and the following scaled goodness of fit indices were examined: Root Mean Squared Error of Approximation (RMSEA), Comparative Fit Index (CFI), Standardised Root Mean Squared Residual (SRMR).

The following suggested diagnostic criterion were used to evaluate the results: CFI > 0.95 **or** SRMR < 0.09. This model fit cut off was based on a combinational rule proposed by Hu & Bentler (1999)[277], taking into consideration the smaller sample size of our study (n < 250). We prioritised a combination of cut offs that would minimise type I error, given our smaller sample size and the existing evidence of structural validity for both PROMs in other populations. We did not consider combinational rules based on RMSEA (or Tucker-Lewis Index (TLI)) as these tend to over-reject both simple and complex true population models in smaller sample sizes.[277]

7.6 Results

Descriptive results

This study included 128 children and young people. Table 7-2 and Table 7-3 show the demographics of those who completed the PIQ-CP and FOPQ-CP, respectively. The number of participants who self-reported for the PIQ-CP was n=115 (Table 7-2), and n=102 for the FOPQ-CP (Table 7-3). Ten items in the PIQ-CP and four in the FOPQ-CP had missing data, making up less than 3% of the sample for each item. As such no items were removed. The PIQ-CP data were positively skewed, with a median [IQR] total score

of 15 [6.0-22.5] and range of 0-48. The FOPQ-CP data were normally distributed with mean (SD) total score of 15.6 (9.3) and range of 0-40.

Table 7-2 Participant demographics of participants who completed the PIQ-CP

Participant characteristic	**PIQ-CP total sample (n=128)**	**PIQ-CP parent report only (n=13)**	**PIQ-CP parent report + self-report (n=14)**	**PIQ-CP self-report (n=115)**	**PIQ-CP Talking Mat (n=43)**	**PIQ-CP pen + paper (n=72)**
Age Mean (SD) Range	13.09 (5.41) 5-29	10.38 (4.96) 5-21	12.2 (5.12) 5-23	13.39 (5.39) 5-29	11.40 (4.68) 5-23	14.58 (5.47) 6-29
Sex						
Male	69 (53.9%)	6 (46.2%)	10 (71.4%)	63 (54.8%)	26 (60.5%)	37 (51.4%)
Female	59 (46.1%)	7 (53.8%)	4 (28.6%)	52 (45.2%)	17 (39.5%)	35 (48.6%)
Administration method						
Talking Mat	43 (33.6%)*	N/A	10 (71.4%)	43 (37.4%)	43 (100%)	N/A
Pen/paper	73 (56.2%)*	N/A	4 (28.6%)	72 (62.6%)	N/A	72 (100%)
Parent report	27 (21.1%)*	13 (100%)	14 (100%)	N/A	10 (23.3%)	4 (5.6%)
Gross Motor Function Classification System						
I	22 (17.2%)	0 (0%)	2 (14.3%)	22 (19.1%)	6 (14.0%)	16 (22.2%)
II	63 (49.2%)	5 (38.5%)	6 (42.9%)	58 (50.4%)	16 (37.2%)	42 (58.3%)
III	13 (10.2%)	0 (0%)	1 (7.1%)	13 (11.3%)	6 (14.0%)	7 (9.7%)
IV	18 (14.1%)	4 (30.8%)	1 (7.1%)	14 (12.2%)	9 (20.9%)	5 (6.9%)
V	12 (9.4%)	4 (30.8%)	4 (28.6%)	8 (7.0%)	6 (14.0%)	2 (2.8%)
Communication ability						
Able to report and describe pain independently	98 (76.6%)	1 (7.7%)	6 (42.9%)	97 (84.3%)	26 (60.5%)	71 (98.6%)

Participant characteristic	PIQ-CP total sample (n=128)	PIQ-CP parent report only (n=13)	PIQ-CP parent report + self-report (n=14)	PIQ-CP self-report (n=115)	PIQ-CP Talking Mat (n=43)	PIQ-CP pen + paper (n=72)
Able to report with a device/method	19 (14.8%)	1 (7.7%)	8 (57.1%)	18 (15.7%)	17 (39.5%)	1 (1.4%)
Unable to report	11 (8.6%)	11 (84.6%)	0 (0%)	N/A	N/A	N/A
Communication Function Classification Level						
I	61 (47.7%)	1 (7.7%)	4 (28.6%)	60 (52.2%)	9 (20.9%)	51 (70.8%)
II	33 (25.8%)	0 (0%)	2 (14.3%)	33 (28.7%)	16 (37.2%)	17 (23.6%)
III	19 (14.8%)	2 (15.4%)	4 (28.6%)	17 (14.8%)	13 (30.2%)	4 (5.6%)
IV	14 (10.9%)	9 (69.2%)	4 (28.6%)	5 (4.3%)	5 (11.6%)	0 (0%)
V	1 (0.8%)	1 (7.7%)	0 (0%)	0 (0%)	0 (0%)	0 (0%)
Parent-reported cognitive impairment**						
Unlikely	60 (46.9%)	1 (7.7%)	3 (21.4%)	59 (51.3%)	9 (20.9%)	50 (69.4%)
Likely	68 (53.1%)	12 (92.3%)	11 (78.6%)	56 (48.7%)	34 (79.1%)	22 (30.6%)
Severity of parent-reported cognitive impairment**	n=68	n=12	n=11	n=56	n= 34	n=22
Mild	24 (35.3%)	0 (0.0%)	2 (18.2%)	24 (42.9%)	9 (26.5%)	15 (68.2%)
Moderate	31 (45.6%)	6 (50.0%)	6 (54.5%)	25 (44.6%)	18 (52.9%)	7 (31.8%)
Severe	10 (14.7%)	6 (50.0%)	3 (27.3%)	4 (7.1%)	4 (11.8%)	0 (0.0%)
Unsure	3 (4.4%)	0 (0.0%)	0 (0.0%)	3 (5.4%)	3 (8.8%)	0 (0.0%)
Has experienced ongoing pain						
Yes	112 (87.5%)	9 (69.2%)	14 (100%)	103 (89.6%)	39 (90.7%)	64 (88.9%)
No	10 (7.8%)	1 (7.7%)	0 (0%)	9 (7.8%)	1 (2.3%)	8 (11.1%)

Participant characteristic	PIQ-CP total sample (n=128)	PIQ-CP parent report only (n=13)	PIQ-CP parent report + self-report (n=14)	PIQ-CP self-report (n=115)	PIQ-CP Talking Mat (n=43)	PIQ-CP pen + paper (n=72)
Unsure	6 (4.7%)	3 (23.1%)	0 (0%)	3 (2.6%)	3 (7.0%)	0 (0%)
Predominant motor type						
Spasticity	82 (64.1%)	5 (38.5%)	8 (57.1%)	77 (66.9%)	24 (55.8%)	53 (73.6%)
Dyskinesia	10 (7.8%)	1 (7.7%)	2 (14.3%)	9 (7.8%)	7 (16.3%)	2 (2.8%)
Mixed	36 (28.1%)	7 (53.8%)	4 (28.6%)	29 (25.2%)	12 (27.9%)	17 (23.6%)

*14 participants completed both parent-report and self-report measures

** Cognitive impairment was determined by asking the young person/caregiver if a formal cognitive assessment had been undertaken. If yes, the result was recorded. If no, the caregiver was asked to suggest a level of cognitive impairment for the young person (none, mild, moderate, severe, unsure)

Table 7-3 Participant demographics of participants who completed the FOPQ-CP

Participant characteristic	FOPQ-CP total sample (n=102)	Talking mat – FOPQ-CP (n=42)	Pen/paper – FOPQ-CP (n=60)
Age mean (SD) Range	13.51 (5.53) 5-29	11.40 (5.05) 5-26	14.98 (5.42) (6-29)
Sex			
Male	54 (52.9%)	21 (50%)	33 (55%)
Female	48 (47.1%)	21 (50%)	27 (45%)
Administration method			
Talking Mat	42 (41.2%)	42 (100%)	N/A
Pen/paper	60 (58.8%)	N/A	60 (100%)
Parent report	N/A	N/A	N/A
Gross Motor Function Classification System (GMFCS)			
I	21 (20.6%)	8 (19%)	13 (21.7%)
II	53 (52.0%)	14 (33.3%)	39 (65.0%)
III	11 (10.8%)	8 (19%)	3 (5.0%)
IV	10 (9.8%)	7 (16.7%)	3 (5.0%)
V	7 (6.9%)	5 (11.9%)	2 (3.3%)
Communication ability			
Able to report and describe pain independently	91 (89.2%)	32 (76.2%)	59 (98.3%)

Participant characteristic	FOPQ-CP total sample (n=102)	Talking mat – FOPQ-CP (n=42)	Pen/paper – FOPQ-CP (n=60)
Able to report and describe pain with the use of a communication device or other method	11 (10.8%)	10 (23.8%)	1 (1.7%)
Unable to report or describe pain	N/A	0 (0%)	0 (0%)
Communication Function Classification Level			
I	57 (55.9%)	18 (42.9%)	39 (65.0%)
II	30 (29.4%)	13 (31.0%)	17 (28.3%)
III	13 (12.7%)	9 (21.4%)	4 (6.7%)
IV	2 (2.0%)	2 (4.8%)	0 (0%)
V	N/A	0 (0%)	0 (0%)
Parent-reported cognitive impairment**			
Unlikely	57 (55.9%)	18 (42.9%)	39 (65.5%)
Likely	45 (44.1%)	24 (57.1%)	21 (35.0%)
Severity of parent-reported cognitive impairment**	n=43	n=18	n=21
Mild	21 (46.7%)	8 (33.3%)	13 (61.9%)
Moderate	18 (40.0%)	10 (41.7%)	8 (38.1%)
Severe	3 (6.7%)	3 (12.5%)	0 (0%)
Unsure	3 (6.7%)	3 (12.5%)	0 (0%)
Has experienced ongoing, everyday pain			
Yes	91 (89.2%)	39 (92.9%)	52 (86.7%)
No	9 (8.8%)	1 (2.4%)	8 (13.3%)
Unsure	2 (2.0%)	2 (4.8%)	0 (0%)
Predominant motor type			
Spasticity	71 (69.6%)	26 (61.9%)	45 (75%)
Dyskinesia	4 (3.9%)	4 (9.5%)	0 (0%)
Mixed	27 (26.5%)	12 (28.6%)	15 (25%)

** Cognitive impairment was determined by asking the young person/caregiver if a formal cognitive assessment had been undertaken. If yes, the result was recorded. If no, the caregiver was asked to suggest a level of cognitive impairment for the young person (none, mild, moderate, severe, unsure)

Structural validity

The results of the CFA for the PIQ-CP and FOPQ-CP are reported in Table 7-4. Structural validity was demonstrated using the proposed one-factor structure for the PIQ-CP (α = 0.89) (Figure 7-2) and the two-factor structure for the FOPQ-CP (fear of pain (α = 0.68) and avoidance of activities (α = 0.80)) (Figure 7-3).

Table 7-4 Confirmatory Factor Analysis findings

Criterion	PIQ-CP – one factor (n=112)	FOPQ-CP – two factor (n=99)
CFI scaled	0.964	0.937
RMSEA scaled	0.086	0.102
SRMR	0.078	0.087

Abbreviations: *CFI = Comparative Fit Index, RMSEA = Root Mean Squared Error of Approximation, SRMR = Standardised Root Mean Squared Residual*

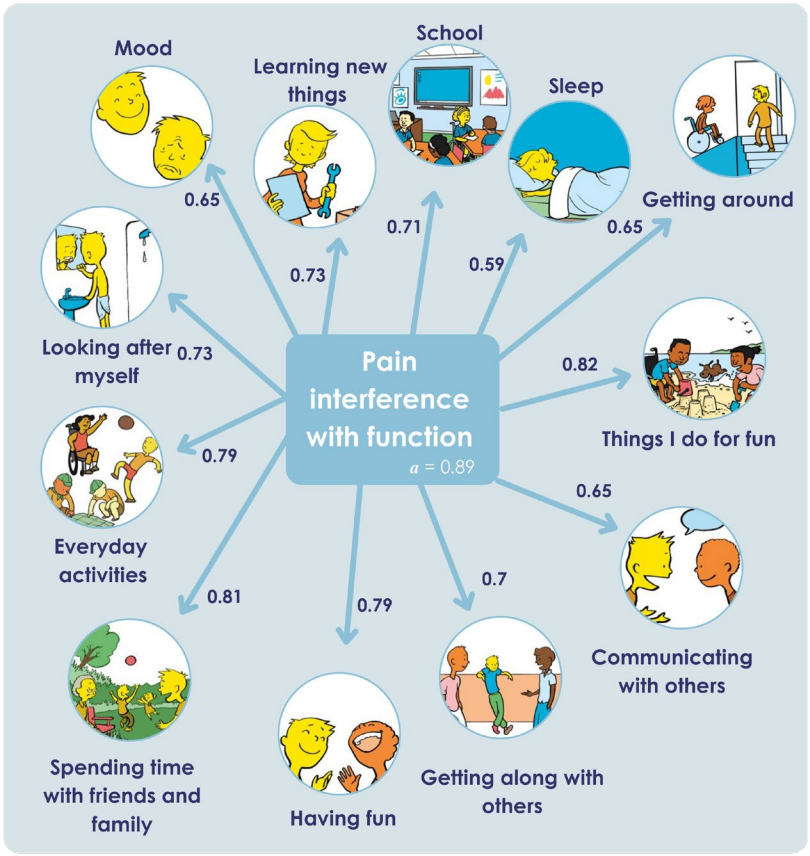


Figure 7-2 CFA findings for the PIQ-CP

Numbers adjacent to the arrows represent the item loading on the factor.
***Abbreviations**: CFA = confirmatory factor analysis, PIQ-CP = Pain Interference Questionnaire for Cerebral Palsy*

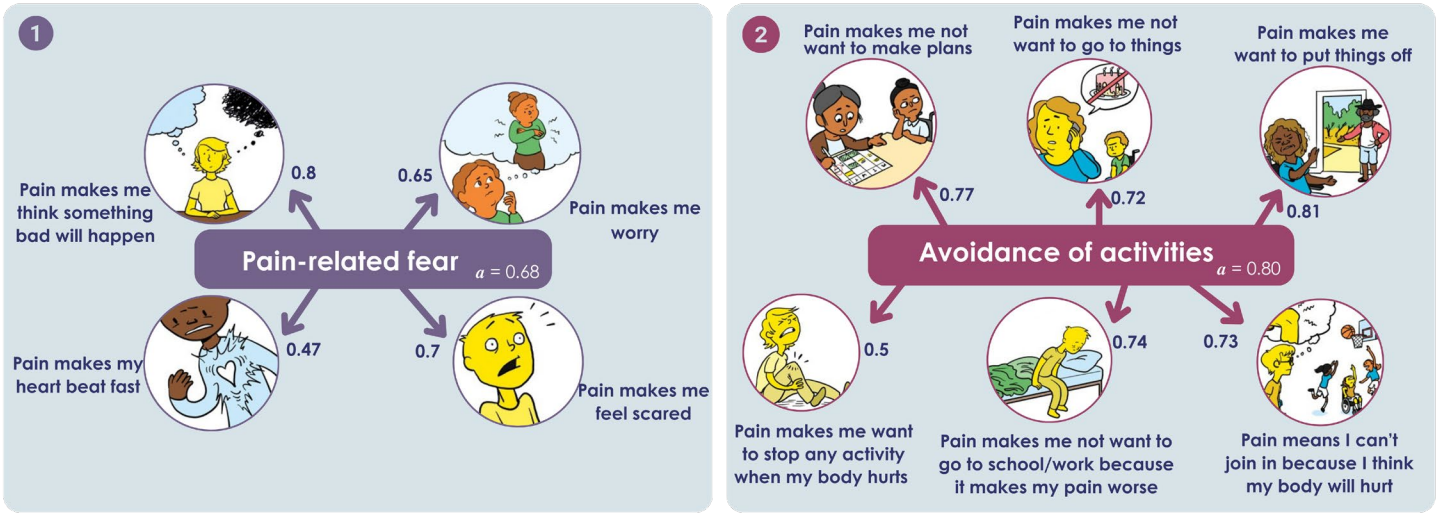


Figure 7-3 CFA results for the FOPQ-CP two factor structure

Numbers adjacent to the arrows represent the item loading on the factor.
***Abbreviations**: CFA = confirmatory factor analysis, F1 = factor 1 (fear of pain), F2 = factor 2 (avoidance of activities), FOPQ-CP = Fear of Pain Questionnaire for Cerebral Palsy*

Ability to self-report

PIQ-CP self-report assessments were completed by 78.9% of all participants (n=101), with a further 10.9% (n=14) completing the self-report in conjunction with parent-report due to uncertainty regarding self-report accuracy. 10.2% (n=13) required a parent-report version only. The demographics of those who required parent-report are provided in Table 7-2. Participants who required parent-report were predominantly CFCS levels IV and V (not always or seldom able to communicate effectively even with familiar people), with moderate to severe cognitive impairment. Of those who completed the PIQ-CP self-report, pen/paper was used by 62.6% (n=72) and *Talking Mats*® by 37.4% (n=43). The FOPQ-CP (self-report only) was completed by 79.6% (n=102) of all participants, with pen/paper used by 58.8% (n=60) and *Talking Mats*® by 41.2% (n=42). The *Talking Mats*® administration method was selected more frequently than pen/paper by participants classified as GMFCS levels IV and V (i.e. primarily non ambulant), CFCS levels III and V (i.e. those who have difficulty communicating with unfamiliar partners), those who require augmentative and alternative communication (AAC) and those with a moderate-severe cognitive impairment.

7.7 Discussion

In this study the PIQ-CP and FOPQ-CP demonstrated sufficient structural validity for children and young people with CP. The adapted tools exhibited the same factor structure as the original tools; a one-factor structure for the PIQ-CP and a two-factor structure for the FOPQ-CP. Furthermore, nearly 80% of children and young people with CP were able to provide self-report using the PIQ-CP and FOPQ-CP. Acceptable internal consistency was reported for the PIQ-CP and the avoidance of activities subscale for the FOPQ-CP, with the fear of pain subscale of the FOPQ-CP marginally below the acceptable range

After establishing adequate content validity of a PROM, the next step in understanding the structure of a reflective PROM is to determine whether or not it has sufficient structural validity.[244] In situations where the model fit is poor, items may sometimes be removed purely to enhance the model's fit. However, this step should not be taken lightly. The cut-offs used in this study (including the use of scaled indices) were based on widely-used and cited literature[277], taking into consideration the comparatively

smaller sample size. As both the PIQ-CP and FOPQ-CP demonstrated sufficient structural validity, no additional evaluation of model fit was required.

Once structural validity has been determined, internal consistency of each subscale can be interpreted.[244] Internal consistency considers how well the items belong together in a scale and whether or not it would be valid to use a sum score, with the COSMIN guidelines suggesting a rule of thumb of α ≥0.70 to demonstrate acceptable internal consistency.[244] In this study, the PIQ-CP (α = 0.89) and the avoidance of activities subscale of the FOPQ-CP (α = 0.80) were above this acceptable range. Although the fear of pain subscale of the FOPQ-CP was marginally below this threshold (α = 0.68), this may reflect its smaller number of items (n=4). Importantly, the Cronbach's alpha 'acceptability' range is a rule of thumb rather than a strict cutoff, and internal consistency should be considered alongside the scale's intended use and the particular population of interest.[278]

This study has shown that the PIQ-CP and FOPQ-CP probably support self-report opportunity for children and young people with CP and with diverse communication and cognitive abilities. The diversity of the ability and age-range of the participants included in this study means the validation findings can be applied to the wider CP population, not just those with typical cognitive and communication abilities. Previous research into chronic pain in CP has been limited by a lack of accessible assessment tools, requiring studies to either exclude participants with diverse abilities, or rely on proxy-reported information.[279] In this study, almost half of the participants likely had some degree of cognitive impairment, which is representative of the CP population.[57] However, up to 80% of the participants in this study were able to complete the self-report measures. This represents a significant improvement compared to previous studies in CP and pain, where fewer than 46% of participants were able to self-report using existing measures such as the PROMIS Pediatric Pain Interference Scale.[12] Furthermore, the participants who were unable to self-report using the adapted tools were primarily classified within CFCS levels IV and V, where individuals are seldom or inconsistently able to communicate even with familiar communication partners. It is therefore understandable they were unable to provide self-report.[16] A small number of participants (n=7) required parent report even though they appeared to have adequate cognitive and communication ability to participate. Anecdotal feedback from clinicians

suggested this was primarily due to co-existing behavioural issues or developmental disabilities affecting behaviour and/or concentration.

This study had some limitations. The cognitive impairment demographic data was based solely on parent-report, without standardised measures such as Intelligence Quotient (IQ). While parent-reported cognitive impairment is not always a precise indicator, given the challenges in using standardised intelligence assessments with children with neurodevelopmental disability[280], we believe the inclusion of formal assessments would not have substantially improved our understanding of this population. In clinical practice, allied health professionals responsible for assessing chronic pain, such as physiotherapists, may not have access to a client's formal cognitive assessment results. Therefore, using a decision-making tree like Figure 1, alongside CFCS level, may be a more practical way to determine a person's capacity to self-report.

Although the sample size met the minimum requirements recommended by COSMIN[244], the accuracy of CFA increases with larger sample sizes and as such it may be useful to conduct a further CFA on a larger sample in the future. The sample size was also slightly skewed towards younger age groups. This reflected the client demographics at the study sites, with only one of the three locations providing services to adult clients. The sample size was also skewed towards ambulant children and young people, however this is consistent with the presentation of CP in the Australian context.[36] Finally, the sample consisted of children and young people with CP attending clinics or accessing therapy services, who may have been more likely to experience pain compared to the general CP population.

In clinical settings, the PIQ-CP and FOPQ-CP can now be used to assess pain interference and pain-related fear/activity avoidance for children and young people with CP, including those with diverse cognitive and communication abilities. Where the clinician and caregiver are unsure of the accuracy of the self-report, it is recommended to complete a parent-report version of the PIQ-CP alongside self-report. Using the PIQ-CP and FOPQ-CP to better understand pain interference and pain-related fear in children and young people with CP may assist in informing efforts to improve access to best practice pain interventions or develop better management strategies. Further investigation of other psychometric properties of the PIQ-CP and FOPQ-CP beyond

structural validity and internal consistency is also strongly recommended. Investigating the test-retest reliability and construct validity of the tools could be beneficial in ensuring reliable use in intervention and longitudinal studies.

7.8 Conclusion

The PIQ-CP and FOPQ-CP have both demonstrated sufficient evidence of structural validity in children and young people with CP. The tools also likely improved self-report opportunity for those with diverse cognitive and communication abilities, allowing most people with CP in this study to self-report. The PIQ-CP and FOPQ-CP can now be used in research and clinical settings to improve understanding of the impact of chronic pain on children and young people with CP, with the longer-term aim of improving access to chronic pain interventions. Future chronic pain research should prioritise self-reported pain assessment by children and young people with CP, including those with diverse cognitive and communication abilities.

Chapter 8 – Discussion and Conclusions

Accurately assessing the impact of chronic pain on all children and young people with CP, regardless of their communication or cognitive abilities, is complex. This thesis sought to address a critical gap in how the impact of chronic pain is assessed in this population. Additionally, this thesis focussed on inclusive and accurate pain assessment as a crucial step toward improving pain management, and subsequently quality of life, for those with CP. This was addressed by identifying, modifying and validating two existing tools for assessing chronic pain in children and young people with CP, with a focus on ensuring accessibility for those with diverse cognitive and communication abilities. Five independent but connected studies, with each individual study building on the previous one, were conducted. Briefly, Study 1 evaluated tools available for assessing pain interference in children and young adults with CP who cannot self-report. Studies 2 and 3 selected and modified existing tools for assessing the impact of chronic pain on emotional functioning for children and young people with CP. Study 4 tested the newly modified tools to ensure they were feasible and comprehensible for children and young people with CP. Finally, Study 5 investigated the psychometric properties of the newly modified tools by using them in practice. My broader PhD project, as well as each individual study, was co-designed with an advisory group of people with lived experience of CP to ensure the research was relevant, accessible and focused on the priorities of people with CP. People with lived experience of CP and their families were also included as research participants in each individual study, prioritising diversity in age as well as cognitive, communication and motor abilities. This was crucial for improving the availability and accessibility of pain assessment tools for **all** children and young people with CP, particularly enabling those who have historically relied on others to report the impact of their pain to now self-report their own experiences.

This final chapter comprises four main sections. Firstly, an integrated summary of the key findings from each chapter are presented (Section 8.1) and secondly, the clinical and research implications of thesis findings are explored (Section 8.2) Thirdly, the

strengths and limitations of the thesis are discussed (Section 8.3) and finally, possible future directions of this work are proposed (Section 8.4).

8.1 Integrated summary of key findings

Each of the five studies has contributed new knowledge within a novel body of work identifying, modifying and validating existing tools for assessing chronic pain interference with function, including emotional functioning, in children and young people with CP and with diverse cognitive and communication abilities.

The first aim of this thesis was to evaluate tools for assessing chronic pain interference in people who are unable to self-report at all, most likely due to their severe cognitive impairment. In the context of assessing pain in children and young people with CP, this group is most vulnerable. Available pain interference tools for this population were identified and evaluated through a systematic review (Chapter 3, Study 1). One tool was found to be recommended for children and adolescents– the Paediatric Pain Profile (PPP)– as it was specifically developed for children and adolescents with severe cognitive impairment. The PPP met the COSMIN recommendations for use in research and clinical practice by demonstrating sufficient psychometric property evidence and feasibility for clinical settings. My systematic review also noted one tool that, with further research and modification, could be recommended for use in children and adults (modified Brief Pain Inventory). A further two tools also had potential for use in the adult population, however these were not explored further in this thesis. While the population included in the systematic review consisted of those unable to self-report, it became clear that many children and young people with CP who have communication and cognitive limitations were previously not given the opportunity to self-report because the available tools did not accommodate their needs. Given this critical gap, and that the first study had successfully answered Aim 1, the subsequent focus of this thesis shifted to adapting pain assessment tools for children and young people with CP who could self-report if tools were modified to support their communication and cognitive abilities. Furthermore, assessing the impact of pain on emotional functioning was considered a priority domain. This was chosen given the influence of this domain on quality of life, the minimal prior assessment of this domain in the CP population and

the importance of improving access to currently underutilised pain-related psychological interventions for children and young people with CP.

The second aim, to adapt key existing tools for the assessment of chronic pain interference with function, including emotional functioning, in children and young people with cerebral palsy, was addressed in two parts, both involving consumers. Firstly, it was necessary to select the tools most relevant and suitable in their current form for children and young people with CP so they could later be modified and validated. Given the need for tools assessing the impact of pain on emotional functioning, tools were grouped into a) multidimensional tools with at least one item assessing impact of pain on emotional functioning and b) tools with a dedicated scale or subscale assessing the impact of pain on emotional functioning. The top two tools from this consumer-driven process were the modified Brief Pain Inventory (mBPI – multidimensional, primarily assessing pain interference) and the Fear of Pain Questionnaire for Children – Short Form (FOPQ-C-SF – dedicated scale, assessing pain-related fear and activity avoidance) (Chapter 4). The chosen tools also supported the findings of Chapter 3 where the proxy report version of the mBPI was identified as a tool showing promise for children and adults who were unable to self-report. Furthermore, the two tools identified in this thesis were also found to be highest-rated in a separate study led by another colleague and co-author, which sought to reach consensus on all tools for the assessment of chronic pain in CP.[281] This additional study, using an entirely separate group of participants, selected the mBPI for pain interference and the FOPQ-C-SF for pain coping.

The mBPI and FOPQ-C-SF were then explored further to identify modifications needed to improve their relevance, comprehensiveness, comprehensibility and feasibility (n=30 participants) (Chapter 4). A range of potential modifications were suggested including reducing the number of response options, simplifying the wording, using visual aids, considering alternative administration methods such as *Talking Mats®* and ensuring the wording was appropriate for people with disability (Chapter 4). As some of the suggestions were conflicting, I undertook a consensus study to determine which of the proposed modifications were most important to implement (n=44 participants) (Chapter 5). This resulted in 21 required changes to the mBPI and four to the FOPQ-C-SF. To enhance accessibility for individuals with diverse cognitive and communication

abilities, two primary administration methods were recommended: standard pen and paper questionnaires and an adapted approach utilising *Talking Mats®*, an evidence-based visual communication framework (Chapter 6). Modifications to both tools were implemented, with the adapted tools now ready to be tested.

For the second part of Aim 2, I sought to examine the key psychometric properties of the CP-adapted tools in children and young people with CP. To recommend a tool for use, COSMIN advises there should be sufficient evidence of all psychometric properties. However where information on all properties is not available, at least sufficient evidence of the content validity (relevance, comprehensiveness and comprehensibility) of the tool is required.[27] Given the relevance and comprehensiveness of the tools been sufficiently investigated in Chapters 4 and 5, my focus for Chapter 6 was on ensuring the comprehensibility of the tools for children and young people with CP. Evaluating the feasibility of these assessments, given they were being modified for use in both research and clinical practice, was also a priority. The adapted mBPI and FOPQ-C-SF were tested with children and young people with CP, demonstrating feasibility and comprehensibility for most, including those with mild-moderate cognitive impairment and AAC users (Chapter 6). Overall, my study demonstrated sufficient evidence of the comprehensibility of the tools in children and young people with CP and chronic pain, which, combined with Chapters 4 and 5, provided robust evidence of the content validity of the CP-adapted tools. The mBPI and FOPQ-C-SF were subsequently renamed the Pain Interference Questionnaire for Cerebral Palsy (PIQ-CP) and the Fear of Pain Questionnaire for Cerebral Palsy (FOPQ-CP), respectively.

Having established the content validity of the PIQ-CP and FOPQ-CP for children and young people with CP, the final study of this thesis explored other key psychometric properties. According to COSMIN, the next priority after content validity is structural validity.[27] Thus, in Chapter 7, a multi-site cross-sectional study was undertaken to assess the structural validity and internal consistency of the two tools. The PIQ-CP demonstrated sufficient structural validity with a one-factor structure, and acceptable internal consistency. Similarly, the FOPQ-CP showed sufficient structural validity with a two-factor structure and acceptable internal consistency for the 'avoidance of activities' subscale. The 'fear of pain' subscale was only minimally below the acceptable threshold for internal consistency, as defined by COSMIN, which was not

considered problematic. Together with the rigorous content validity process described in Chapters 4-6, these findings support recommending the PIQ-CP and FOPQ-CP for use in practice. However, further research is still needed to investigate remaining psychometric properties, such as test re-test reliability and responsiveness to change.

Overall, this thesis has contributed new knowledge by selecting, modifying and validating two existing chronic pain assessment tools, now known as the PIQ-CP and FOPQ-CP, for children and young people with CP. These are the first tools specifically modified to enable self-reporting of chronic pain interference with function, including emotional functioning, in children and young people with CP, including those with diverse cognitive and communication abilities. The PIQ-CP and FOPQ-CP are clinically feasible and have sufficient evidence of acceptable psychometric properties for use in practice. For those children and adolescents with CP who, even with adaptations, are still unable to self-report, the PPP is recommended.

8.2 Clinical and research implications of thesis findings

8.2.1 Recommendations for how these tools are used in clinical and research settings

The aim throughout this thesis was to identify, modify and validate pain tools which could be feasibly used in usual clinical and research settings with children and young people with CP. This was a priority given some tools have excellent psychometric properties but are not clinically feasible. For example, the NCCPC-R requires two hours of observation of the child or young person.[200] The PIQ-CP and FOPQ-CP are now freely available through the Australasian Academy of Cerebral Palsy and Developmental Medicine (AusACPDM) website, meaning they can be used by anyone in clinical practice or research. *Talking Mats® Ltd* do, however, recommend undertaking *Talking Mats®* training prior to administering the *Talking Mat* versions of the tools. This is important in ensuring the child or young person's views are prioritised and accurately elicited. The training is both easily accessible (online via a website or face to face) and relatively low-cost.

In clinical settings, the PIQ-CP and FOPQ-CP can be used to assess and monitor chronic pain interference and pain-related fear/activity avoidance for children and young people with CP, including those with diverse cognitive and communication

abilities. Where the clinician or caregiver is unsure of the accuracy of the self-report, it is recommended to complete a parent-report version of the PIQ-CP alongside self-report. Both the PIQ-CP and FOPQ-CP are recommended tools within the recently developed chronic pain assessment framework for CP (Smith N, *under review*). It is anticipated regular use of these tools in practice will assist clinicians in identifying children and young people with CP who may benefit from chronic pain interventions, including referral to specialist tertiary chronic pain services.

In research settings, this thesis provides clear guidance on using the PPP for assessing pain interference in children and young people with CP who cannot self-report. Additionally, it recommends using the PIQ-CP and FOPQ-CP to assess pain interference and pain-related fear/activity avoidance, respectively, in children and young people who can be supported to self-report. This will allow more children and young people with CP who have a range of cognitive and communication abilities to provide self-reported information in research studies, where historically they have been excluded or restricted to proxy report.[266] This is crucial as without first comprehensively assessing, it cannot be assumed pain is not an issue. The tools also have the potential to be used to evaluate pre and post intervention change in both clinical and research settings, however further psychometric property investigation is required to understand the reliability and responsiveness to change of the PIQ-CP and FOPQ-CP.

8.2.2 Moving beyond pain intensity when assessing chronic pain in children and young people with CP

This thesis has addressed a gap in assessing chronic pain interference, including impact of pain on emotional functioning, in individuals with CP, moving beyond a limited focus on pain intensity. While pain intensity is important for understanding acute pain, it offers limited insight into how ongoing pain affects a person's life. In the context of chronic pain, intensity can fluctuate and may not directly reflect the extent to which pain limits daily activities or affects emotional functioning.[22] The complexity and diversity within the CP population has made it challenging to assess even pain intensity, which is typically the simplest aspect of pain to measure. This difficulty has been especially pronounced for individuals with diverse communication and cognitive abilities.[112, 113] Consequently, comprehensive chronic pain assessment has often been avoided or limited for many people with CP. The PIQ-CP and FOPQ-CP, developed

through this thesis, provide an accessible way to better understand the impact of chronic pain on a wider group of people with CP, rather than only those who are 'easy' to assess because of their cognitive abilities. This is the first body of work to assist self-report of the impact of chronic pain from people with CP and with diverse cognitive and communication abilities. Better understanding of the impact of pain, reported where possible by the person experiencing it, is the first step in providing equitable access to intervention.

While comprehensive assessment of chronic pain has been limited in both research and clinical practice for the whole CP population,[9, 41] assumptions about what self-report involves have further limited access to self-report measures for those with diverse cognitive and communication abilities. In Chapter 7 (Study 5), I found approximately 80% of children and young people with CP can provide self-report of the impact of pain using the PIQ-CP and FOPQ-CP. For approximately 10%, both the clinician and parent questioned the reliability of the self-report, so a parent report was also completed. This contrasts most patient-reported outcome measures (PROMs) which restrict self-report, including the self-report of pain, to those with the cognitive abilities of a person 8 years and older.[255] Given 50% of children and young people with CP have some form of cognitive impairment[36], restricting self-report only to those with the cognitive ability of an eight-year old is an inequitable method of assessing the impact of chronic pain.[247] While other research has taken steps to provide avenues for self-report in people with cognitive impairment[248], it has not considered the complex interplay of diversity in cognitive, communication **and** motor abilities like those seen in the CP population.

8.2.3 Prioritising self-report for the benefit of people with CP and their families

The studies in this thesis prioritised the self-reporting of pain wherever possible, even for people with diverse communication and cognitive abilities. The United Nations recognises self-report of health-related information, including by using PROMs, as a right for children and people with disabilities. This right ensures individuals actively engage in healthcare decisions about their own lives.[248] For young people with CP who have significant communication and cognitive limitations and are living away from home, there may not be someone who knows them well enough to provide a reliable proxy report. Even for parents of children with CP, providing a proxy report can be

challenging. In Chapter 6, parents often reported learning new information about their child when their child was facilitated to self-report using the PIQ-CP and FOPQ-CP. Although some parents felt confident to report the degree to which pain interfered with their child's life, few felt they could accurately report how their child perceived pain-related fear (Chapter 5). While prioritising self-reported assessment of the impact of chronic pain has clear benefits for the child or young person by 'hearing their voice' and allowing engagement in healthcare decision-making, it also benefits the parents by ensuring they do not have to 'guess' their child's pain. Parents have reported the difficulties associated with proxy-reporting their child's pain can cause them stress and contribute to feeling 'not in control'.[261] Rinde et al. (2024) found, even for children who were verbal, parents were not sure of their own judgements about when their child was in pain and were afraid they may have overlooked something.[261] This was further complicated by parents reporting their child sometimes used pain as an excuse to avoid situations or activities they did not like.[261] However there will always be some children and young people who are not able to self-report due to severe cognitive impairment. In these cases, it is advisable to use a mix of observational and proxy-reported outcome measures to guide parents in reporting their child's pain.[113, 118]

8.2.4 Consumer involvement throughout the research process

Successfully adapting the tools to ensure accessibility for a wider range of children and young people with CP would not have been possible without the integrated engagement of people with CP, health professionals and their families at every stage of the project. This thesis prioritised the inclusion of children and young people with CP and with diverse motor, communication and cognitive abilities throughout the individual studies, using deliberate adaptations to the study methods to ensure they could provide input. A further strength of this thesis was the involvement of people with lived experience in overseeing the project direction and each individual stage. Previous research conducted in chronic pain populations has highlighted a disparity between research topics considered important by researchers, when compared to people with lived experience of the condition.[105, 282] This disparity is thought to be responsible for the limited uptake of research findings, as the research is not meaningful to people with lived experience and their families.[282] To ensure this project was meaningful to people with CP, I focused on a key research priority of pain assessment and management, which was identified in the top five research priorities for people with lived experience of

CP, clinicians and researchers.[104] This project also included two key strategies to ensure relevant and meaningful research: 1) guidance from a lived experience advisory group throughout the project, and 2) prioritising the involvement of people with lived experience with a diverse range of abilities.

The lived experience advisory group was involved from project conception all the way through to completion. The group consisted of two young adults with CP and a parent of a child with CP, and was representative of the diverse motor, communication and cognitive abilities within the CP population. The group met bimonthly throughout the project and provided guidance and recommendations for each stage. Examples included recommending specific research methods, adapting interview procedures, reviewing research findings, assisting with interpretation of research findings, guiding the research focus and suggesting future directions. The group also provided advice on key aspects of the research methodology, for example specifying the included age range for testing the tools (Chapter 6). The advisory group recommended an age range of 5-30 years, which was a broader age range than originally considered. The reason for this recommendation was two-fold. Firstly, to ensure the modifications made to the tools to enhance cognitive accessibility enabled understanding for a person with the cognitive ability of a five-year-old child; and secondly to extend the cut off for young adulthood to 30 years, recognising this period may be extended for individuals with disability due to delayed transitions to adulthood social roles.

The advisory group was recruited via contacts known to me through existing clinical relationships and through the AusACPDM Pain in CP special interest group. These existing relationships proved a strength and likely improved buy-in and continued involvement over the multiple years of the project. The advisory group were reimbursed appropriately for their time to recognise the expertise they provided.

The project, while methodologically based on COSMIN, was also influenced by the principles of evidence-based co-design methodology.[229] The involvement of people with lived experience as research participants and advisors was crucial to the success of this project. Most importantly, participants were representative of the spectrum of motor, communication and cognitive ability seen in the CP population, which required a staged approach to participation. For this reason, I prioritised research methods and co-design procedures that could be adapted to be accessible.[267] The modifications

required to ensure people with diverse abilities could participate in the research was guided by the advisory group and recent research by Walsh and colleagues.[283] Other research has used arts-based participatory methods, such as body mapping and photo voice, to identify the key constructs of interest for patient-reported experience measures (PREMs) for children with intellectual disability in healthcare settings.[267] In the case of this thesis, the construct was already identified, meaning the focus was instead on improving accessibility and understanding of the tools using methods such as *Talking Mats*®.[229] Future pain management research in CP could potentially benefit from using arts-based participatory approaches as part of co-design methodology to consider which interventions may be most appropriate for people with CP.

8.2.5 Development of a PROM modification process which could be used in other populations

When considering the modification process throughout this thesis, several adjustments were made as the COSMIN methodology did not have specific recommendations for the inclusion of people with diverse cognitive and communication abilities. For this reason, the modification process described in this thesis was informed by three key ideas:

1) Developing new tools is a time-consuming process, and COSMIN recommend, where possible, existing PROMs should be tested for content validity in new populations, rather than developing a new tool[31, 196]
2) Existing PROMs are rarely developed in populations with complex communication needs and cognitive impairment
3) It is not possible to test the content validity of tools developed for other populations across the spectrum of ability in CP without first modifying, as the tools are too complex and lack accessibility to even be tested

Therefore, the approach used in this thesis initially modified the tools using participants within the CP population without cognitive impairment, and proxy reporters of those with cognitive/communication impairment (Chapters 4 and 5), so the tools could then be presented for pilot testing in an accessible form (Chapter 6). The standard COSMIN process for an existing PROM is to ask participants to rate the relevance, comprehensiveness and comprehensibility of existing items within an item bank, and then select the most meaningful items for inclusion.[284] This process also considers the age and technology literacy of the population.[285] Given the breadth of cognitive and

communication impairment in the CP population, we needed to include representatives of those with cognitive and communication impairment to first modify the tools so they were then in an accessible format for further testing across the whole target population. This method appeared to be successful as the modified tools were accessible and comprehensible to people with diverse cognitive and communication abilities when tested in Chapter 6. The process outlined in this thesis could be applied to tools assessing constructs other than pain, for example fatigue or mental health.

8.2.6 Knowledge translation

Throughout this thesis, the advisory group and participants with lived experience reiterated the importance of ensuring improved pain assessment leads to improved pain management. It is insufficient to solely assess pain using the PIQ-CP and FOPQ-CP; the information gained from these assessments must prompt clinicians to consider how pain will be better managed. Presently, the limited pain management options available to children and young people with CP makes this process challenging.[9] However, the use of the PIQ-CP and FOPQ-CP in practice may bring the issue of chronic pain to the forefront for clinicians. To begin disseminating knowledge about the PIQ-CP and FOPQ-CP, I have published peer-reviewed publications, presented at national and international conferences both in the pain and disability research fields and presented webinars on freely accessible platforms, including CP-Achieve, AusACPDM Listening & Sharing sessions and the Wired-On Development Clinical Excellence Summit. These platforms strategically target both clinicians and researchers. The wide-spread recruitment necessary for Study 5 (Chapter 7) enabled the provision of educational outreach regarding the newly developed-tools, and the project, to clinicians at three healthcare organisations (Novita, Women's and Children's Hospital and Perth Children's Hospital). My continued clinical work in South Australia has allowed for knowledge dissemination about the project and the importance of pain assessment to other clinicians locally in the CP field. While knowledge implementation outcomes were not formally assessed in this thesis, the anecdotal findings from each study suggest the CP-adapted tools are acceptable, appropriate, feasible and sustainable for use in clinical practice.[282] Furthermore, the tools are now publicly and freely available on the AusACPDM website. The AusACPDM is the peak body in Australia and New Zealand for cerebral palsy and developmental medicine. Furthermore, there has been interest from individuals outside the project team wanting to use the adapted tools themselves in

research and clinical settings. Finally, a complementary but separate PhD project has developed a framework for selecting tools for the assessment of chronic pain in CP in routine clinical practice. The adapted tools from my thesis have been recommended as part of this framework, providing an additional avenue for knowledge translation.

The completed activities from this thesis fit within the first four stages of the Knowledge to Action map, including identifying the problem (Chapters 3 and 4), adapting knowledge to the local context (Chapters 4 and 5), assessing barriers to knowledge use (Chapter 4) and selecting, tailoring and implementing interventions (Chapters 6 and 7).[286] This is a similar initial process to that undertaken by Stewart et al. (2019) in a project aiming to implement accurate assessment of dyskinesia in CP into clinical practice.[287] While barriers to knowledge use were only superficially identified in Chapter 4, one of the likely knowledge translation barriers will be limitations on clinician time in implementing the modified tools into clinical practice, particularly for children and young people with CP and complex communication needs. As discussed in Chapter 6, the time taken to complete the assessments varies widely, with children and young people with complex communication needs and cognitive impairment taking much longer to complete. It is necessary for clinicians to adapt to prioritise self-report of pain, while also balancing the fatigue associated with completing assessments for those with complex communication needs. It is likely the PIQ-CP and FOPQ-CP may be most appropriate and useful as part of a wider assessment, for example as part of surveillance services such as CP-UP in Sweden.[264] These surveillance assessments are designed to consider a person's needs holistically, while also providing recommendations for follow up and intervention. Pain assessments could also be completed separately from a standard clinical appointment, for example with a parent or caregiver at home ahead of time and/or using patient portals in electronic medical records. It is likely the strategies used to ensure implementation into routine clinical care will vary widely between different health services and organisations, depending on the needs of the client. Regardless of the strategies used, it is of utmost importance that the impact of pain is routinely assessed in clinical practice.

8.3 Limitations

This thesis has several potential limitations which need to be considered when interpreting the results collectively. Firstly, the study population captured within this thesis was primarily geographically located within Australia and New Zealand. Secondly, the comprehensibility testing of the adapted tools was carried out on children and young people with CP within Australia only. Further work is required to investigate the cross-cultural validity of the adapted tools, ensuring they are similarly comprehensible, comprehensive and relevant to children and young people with CP in other countries, particularly non-English speaking countries.[27] Thirdly, the included participants in this thesis were reflective of the highly heterogenous CP population in terms of motor, cognitive and communication abilities, which is both a strength and a limitation. The inclusion of such a diverse group ensured the clinical feasibility of the tools, however it also highlighted the need to be adaptable and flexible when administering the assessments. For example, while some PROMs stipulate exact wording and administration conditions, the diversity of need and ability in the CP population led us to develop an administration guide with flexibility for different levels of support. The administration guide also provides examples of how to describe 'pain', as it was identified not all children and young people with CP understand what pain is, as it is a usual part of their life.[9, 211, 261]

When modifying the tools, it was not always possible to implement known features of cognitive accessibility, such as those developed by Kramer and colleagues.[248] The focus of this thesis was on adapting existing tools and it was important to ensure the construct of interest and individual item meaning was preserved through the modification process. While many of the modifications to the tools were consistent with several features of cognitive accessibility, including consistent layout, use of font and colour and visual integration, it was not always possible to use positive wording.[248] For a concept such as pain-related fear/avoidance, positive framing for some of the items may have confused the inherently negative construct. This is not, however, considered a significant limitation as this thesis triangulated literature recommendations on cognitive accessibility, a qualitative study and a consensus study when determining appropriate modifications. This likely contributed to the ease of implementation of the tools into practice in Chapter 6, with only minor changes required at the end of the pilot testing process. In contrast, the cognitive accessibility recommendations by Kramer and

colleagues had been developed from a literature review only, and did not involve consumers, or gain consensus on the most relevant features.[248] A further potential limitation was when considering the abstract constructs of pain-related fear and activity avoidance (FOPQ-CP). It was only possible to simplify to a certain degree due to the complexity of the construct. After modification, the constructs of pain-related fear and activity avoidance remained complex and abstract. However, these constructs were still deemed important to assess, given the connection between pain-related fear and activity avoidance and the adverse effects of chronic pain.[26, 111]

The resource and time constraints of a PhD meant it was not possible to modify additional PROMs beyond the top-rated tools identified in Chapter 4. Work completed by Smith N et al. (under review) identified a range of PROMs which could be used to assess the impact of chronic pain on children and young people with CP, of which the original mBPI and FOPQ-C-SF were recommended. Future work could consider using a similar modification process to the one followed in this thesis to modify additional tools, ensuring broader assessment of chronic pain. Of particular value would be modifying a pain identification tool, such as the Pediatric Pain Screening Tool suggested by Smith N et al.[281], to allow simple and quick detection of pain in busy clinic settings.

This thesis did not explore electronic versions of the two modified assessment tools even though electronic PROMs (ePROMs) are known to reduce administration times and improve clinical decision making and symptom management.[285] This decision was influenced by four factors: 1) the advice of the consumer advisory group, 2) the ability to integrate an ePROM with multiple different electronic medical record (EMR) systems across the study sites, 3) the accessibility and flexibility of ePROMs for people with complex communication needs and cognitive impairment and 4) the resource constraints of the project. Of the three sites involved in Study 5 (Chapter 7), one did not have an EMR system and the remaining two sites had different EMR systems, neither of which had a patient-facing portal in which to administer ePROMs. There were also concerns from the advisory group and the project team about the accessibility of electronic versions of the assessments for people with diverse cognitive and communication abilities. For example, the Cerebral Palsy Research Network community registry in the United States has implemented regular pain assessment through online surveys for adults.[288] However the diversity of participants self-reporting

in these assessments has been limited to those who have the cognitive ability to participate independently online. This highlights some of the challenges of ePROM distribution and the need to consider those who need adaptation or in-person support to participate. It also raises the question of the most effective and inclusive way to use ePROMs in the CP population, while also ensuring sustainability of the digital platform.

Furthermore, given the time and financial constraints of this PhD project, pursuing an ePROM option was not feasible, especially given the significant customisation required to ensure accessibility across the population. I initially explored the possibility of using an existing paediatric ePROM platform, Voxe[289, 290], however this was not possible as the associated international cloud storage did not meet the local Human Research Ethics Committee (HREC) requirements. In the future, ePROM versions of the PIQ-CP and FOPQ-CP could potentially be integrated with an individual's AAC device, enabling greater customisation. However, the long-term sustainability and maintenance requirements of an ePROM platform should be carefully considered, particularly given the potential impact of usage costs on public accessibility.[291]

8.4 Future directions

The findings reported in this thesis suggest several promising directions for future research. This section will explore three key areas for further investigation, including further psychometric property investigation for the adapted tools, improving our understanding of the impact of pain in CP and enhancing pain management for all children and young people with CP.

8.4.1 Further psychometric property investigation

The tools modified in this thesis have demonstrated sufficient evidence for content validity, the most important psychometric property for selecting a PROM in practice.[27] However, sufficient evidence across all psychometric properties is necessary to ensure accurate assessment in all contexts.[244] As I have only investigated these tools in an Australian context, it would be beneficial to investigate the cross-cultural validity or measurement invariance of the CP-adapted tools in different cultural and language speaking groups, or to investigate the impact of age on responses to different items.[244] If the tools are to be used as part of longitudinal studies or clinical trials, it is necessary

to investigate responsiveness to change.[244] The original versions of the tools both had sufficient evidence for construct validity, which is important as investigating the construct validity of the modified tools may pose a challenge.[154, 222] This is because no other pain interference tools use modified administration methods like those used for the PIQ-CP and FOPQ-CP presented in this thesis. As such, assessing correlations between the PIQ-CP and FOPQ-CP and other measures may be difficult, as other measures are not accessible to all individuals with CP.

8.4.2 Improved understanding of pain interference and pain-related fear in CP

The logical next step is to use the PIQ-CP and FOPQ-CP to better understand the profile of pain-related fear and pain interference in children and young people with CP with diverse cognitive and communication abilities. Children and young people with CP may cope with and think about pain differently to those who do not have CP, and gathering this information will influence how interventions are developed or modified to better suit children and young people with CP.[292] While other research has reported pain-related fear and coping strategies for children and young people with CP, those with cognitive impairment or complex communication needs have been excluded.[292] Young people with diverse abilities should be included in future studies to ensure interventions are tailored to the needs of all children and young people with CP and to ensure equitable participation in research. Better understanding of the impact of pain in CP may identify the need for improved access to existing chronic pain interventions, or help to inform the development or adaptation of pain interventions specific for children and young people with CP.

8.4.3 Enhancing pain management in CP

Given the advances made through this thesis in chronic pain assessment for children and young people with CP, and the work of Smith N et al.[113], it is imperative the research focus now shifts to how chronic pain can be better managed in this population. While further refinements may be needed for chronic pain assessment tools, exploring, designing and adapting interventions is a critical priority.[9] I propose three key areas for pain management research in CP, informed by Harvey and colleagues (2024)[293]: 1) adapting existing chronic pain management interventions, 2) improving the uptake of psychological strategies for managing pain and 3) supporting children and families to cope with and better self-manage chronic pain.

While the gold standard of interdisciplinary care for chronic pain management is well established[293], children and young people with CP do not have ready access to these existing services.[19, 102] As a first step, identifying barriers to accessing existing tertiary specialist pain management services for children and young people with CP, and for clinicians providing those services, would be beneficial. The interdisciplinary model of care recommended for chronic pain management is similar to the one currently used for children and young people with CP to manage their physical disability. This similarity suggests there are opportunities to expand and enhance the existing model to incorporate chronic pain management. It would also be beneficial to investigate ways to adapt pain management interventions for children and young adults with diverse cognitive and communication abilities, alongside developing specific pain-reducing movement programs for those with significant mobility limitations who find it difficult to engage in movement in the home and community environments.[294]

Much work has already been done in developing pain education resources for children, such as the picture books 'Zoe and Zak's Pain Hacks', for adolescents[295, 296] (Young Pain Health website, ICanCope app & the Power over Pain Portal – youth) and for young adults without disability[297] (Pain Health website & the Power over Pain Portal - adult). Without re-inventing the wheel, existing resources could be adapted through co-design to be more appropriate and accessible for children and young people with CP and with diverse abilities. This may require a similar approach to that undertaken in this thesis, in which the resources are initially modified into a more accessible format and then tested more widely in the population. Furthermore, considering how to improve the uptake of psychological strategies for managing pain, such as strategies to improve pain coping for children and young people with CP, is a priority.[298] This could be delivered through increased access to psychological services, via a pain education platform, by upskilling existing members of the child or young person's interdisciplinary team, or by upskilling parents.[298] Combining a range of strategies (i.e. physical, psychological, pharmacological and preventative strategies) for managing pain, implemented by a child's or young person's multidisciplinary team and family, would likely be beneficial.[298]

Finally, it is important to consider the child and young person's pain experience in the context of their family.[299] Future work should support families and caregivers, alongside

children and young people with CP, to better cope with and manage pain. The impact of chronic pain on the wider family is well-documented; parental mental health problems are common among children with chronic pain[261, 300] and adherence to pain treatment regimens is heavily influenced by the parent's experience of their child's treatment.[301] This is likely to have an even greater impact on parents of children and young people with CP, who are already involved in managing complex treatment regimens for their child's physical disability. Emerging research also highlights the importance of families validating their child's pain to promote better chronic pain outcomes.[298] This is especially significant for marginalised groups, such as children and young people with CP.[302]

8.5 Conclusions

This thesis has contributed important new knowledge through the selection, modification and validation of two chronic pain assessment tools, now known as the PIQ-CP and FOPQ-CP, for children and young people with CP. These are the first tools specifically modified to enable self-reporting of chronic pain interference with function, including emotional functioning, in children and young people with CP, including those with diverse cognitive and communication abilities. The PIQ-CP and FOPQ-CP are clinically feasible, freely available and have sufficient psychometric property evidence for use in practice. These tools address gaps in current assessment practices, enabling more equitable evaluation of the impact of chronic pain on children and young people with CP, who might otherwise have been overlooked. For those children and adolescents with CP who, even with adaptations, are still unable to self-report, the PPP is recommended. Future research should now focus on using these CP-adapted tools to better understand the impact of chronic pain on all children and young people with CP, in order to improve access to pain management interventions.

Appendices

Appendix A: CP-adapted pain assessment tools

These are now publicly available, hosted on the AusACPDM website via Figshare.

Appendix A1: Pain Interference Questionnaire for Cerebral Palsy – pen/paper self-report

Appendix A2: Pain Interference Questionnaire for Cerebral Palsy – parent report

Appendix A3: Pain Interference Questionnaire for Cerebral Palsy – *Talking Mats®*

Appendix A4: Pain Interference Questionnaire for Cerebral Palsy – administration guide

Appendix A5: Fear of Pain Questionnaire for Cerebral Palsy – pen/paper

Appendix A6: Fear of Pain Questionnaire for Cerebral Palsy – *Talking Mats®*

Appendix A7: Fear of Pain Questionnaire for Cerebral Palsy – administration guide

Appendix B: Supplementary material from Chapter 3

Links to individual appendices are provided below. All appendices related to Chapter 3 are freely available in the 'Supporting information' section of the open-access published article: https://doi.org/10.1111/dmcn.15535.

Appendix B1: Search strategies

Appendix B2: PRISMA flow diagram for studies identified via searches of databases and citation searching

Appendix B3: Characteristics of included studies

Appendix B4: Characteristics of pain interference assessment tools for individuals unable to self-report

Appendix B5: Reasons for exclusion of tools

Appendix B6: Studies assessing psychometric properties of translations of included tools

Appendix B7: Summary/pooled result for each psychometric property

Appendix B8: Risk of bias scores

Appendix B9: PROMIS PPPIS measurement properties for computer adaptive test, item bank, fixed short form and custom short form

Appendix C: Supplementary material from Chapter 4

Links to individual appendices are provided below. All appendices related to Chapter 4 are freely available in the 'Electronic supplementary material' section of the open-access published article: https://doi.org/10.1007/s11136-024-03693-1

Appendix C1: Decision making re: inclusion of tools in online survey

Appendix C2: Survey

Appendix C3: Survey questions and their relevance to the content validity and feasibility domains:

Appendix C4: Interview guide

Appendix C5: Codebook and refined coding schema

Appendix C6: Survey ratings

Appendix C7: Modification suggestions mapped to big picture categories

Appendix C8: Direct quotes and their category mapping

Appendix D: Supplementary material from Chapter 5

Links to individual appendices are provided below. All appendices related to Chapter 5 are freely available in the 'Supplemental Digital Content' section of the open-access published article: https://doi.org/10.1097/PR9.0000000000001304

Appendix D1: Suggested changes from qualitative descriptive study grouped into those to be presented in the Delphi study, those that do not need Delphi consensus and those not to be presented

Appendix D2: Delphi round 1

Appendix D3: Delphi round 2

Appendix E: Supplementary material from Chapter 6

Links to individual appendices are provided below. All appendices related to Chapter 6 are freely available in the 'Electronic supplementary material' section of the open-access published article: https://doi.org/10.1007/s11136-025-03981-4

Appendix E1: Interviewer notes template

Appendix E2: Interview guide

Appendix E3: Cognitive interviewing interpretation of items and changes made

Appendix E4: Participant characteristics per version and tool

Appendix E5: direct quotes and their category mapping

Appendix E6: Suggested changes

Appendix F: Supplementary material from Chapter 7

Appendix F1: Suggested factor structure based on previous CFA in the original population for the FOPQ-C-SF

Fear of Pain (Factor 1)	Avoidance of Activities (Factor 2)
Pain makes my heart beat fast	Pain makes me not want to go to things
Pain makes me feel scared	Pain makes me want to stop any activity when my body hurts
Pain makes me worry	Pain makes me not want to go to school/work because it makes my pain worse
Pain makes me think something bad will happen	Pain makes me not want to make plans
	Pain makes me want to put things off
	Pain means I can't join in because I think my body will hurt

References

1. McIntyre S, Goldsmith S, Webb A, Ehlinger V, Hollung SJ, McConnell K, et al. Global prevalence of cerebral palsy: A systematic analysis. *Dev Med Child Neurol*. 2022; **64**(12): 1494-506.
2. Rosenbaum P, Paneth N, Leviton A, Goldstein M, Bax M, Damiano D, et al. A report: the definition and classification of cerebral palsy April 2006. *Dev Med Child Neurol Suppl*. 2007; **109**: 8-14.
3. McKinnon CT, Meehan EM, Harvey AR, Antolovich GC, Morgan PE. Prevalence and characteristics of pain in children and young adults with cerebral palsy: a systematic review. *Dev Med Child Neurol*. 2019; **61**(3): 305-14.
4. Vega R, Groenewald C, Bromberg MH, Beals-Erickson SE, Palermo TM. Chronic pain prevalence and associated factors in adolescents with and without physical disabilities. *Developmental Medicine And Child Neurology*. 2018; **60**(6): 596-601.
5. Russo RN, Miller MD, Haan E, Cameron ID, Crotty M. Pain characteristics and their association with quality of life and self-concept in children with hemiplegic cerebral palsy identified from a population register. *Clin J Pain*. 2008; **24**(4): 335-42.
6. Shearer HM, Côté P, Hogg-Johnson S, McKeever P, Fehlings DL. Pain trajectories and well-being in children and young people with cerebral palsy: A cohort study. *Developmental Medicine And Child Neurology*. 2022.
7. Østergaard CS, Pedersen NSA, Thomasen A, Mechlenburg I, Nordbye-Nielsen K. Pain is frequent in children with cerebral palsy and negatively affects physical activity and participation. *Acta Paediatrica*. 2021; **110**(1): 301-6.
8. Westbom L, Rimstedt A, Nordmark E. Assessments of pain in children and adolescents with cerebral palsy: a retrospective population-based registry study. *Developmental Medicine & Child Neurology*. 2017; **59**(8): 858-63.
9. Harvey A, Smith N, Smith M, Ostojic K, Berryman C. Chronic pain in children and young people with cerebral palsy: a narrative review of challenges, advances, and future directions. *BMC Med*. 2024; **22**(1): 238.
10. Doralp S, Bartlett DJ. The Prevalence, Distribution, and Effect of Pain Among Adolescents with Cerebral Palsy. *Pediatric Physical Therapy*. 2010; **22**(1).
11. Parkinson KN, Dickinson HO, Arnaud C, Lyons A, Colver A. Pain in young people aged 13 to 17 years with cerebral palsy: cross-sectional, multicentre European study. *Archives of Disease in Childhood*. 2013; **98**(6): 434.
12. Ostojic K, Paget S, Kyriagis M, Morrow A. Acute and Chronic Pain in Children and Adolescents With Cerebral Palsy: Prevalence, Interference, and Management. *Arch Phys Med Rehabil*. 2020; **101**(2): 213-9.
13. Penner M, Xie WY, Binepal N, Switzer L, Fehlings D. Characteristics of Pain in Children and Youth With Cerebral Palsy. *Pediatrics*. 2013; **132**(2): e407.
14. Palisano RJ, Rosenbaum P, Bartlett D, Livingston MH. Content validity of the expanded and revised Gross Motor Function Classification System. *Dev Med Child Neurol*. 2008; **50**(10): 744-50.
15. Eliasson A-C, Krumlinde-Sundholm L, Rösblad B, Beckung E, Arner M, Öhrvall A-M, Rosenbaum P. The Manual Ability Classification System (MACS) for children with cerebral palsy: scale development and evidence of validity and reliability. *Developmental Medicine And Child Neurology*. 2006; **48**(7): 549-54.
16. Hidecker MJC, Paneth N, Rosenbaum PL, Kent RD, Lillie J, Eulenberg JB, et al. Developing and validating the Communication Function Classification System for individuals with cerebral palsy. *Dev Med Child Neurol*. 2011; **53**(8): 704-10.

17. Sellers D, Mandy A, Pennington L, Hankins M, Morris C. Development and reliability of a system to classify the eating and drinking ability of people with cerebral palsy. *Developmental Medicine And Child Neurology*. 2014; **56**(3): 245-51.
18. Lee RR, Rashid A, Ghio D, Thomson W, Cordingley L. Chronic Pain Assessments in Children and Adolescents: A Systematic Literature Review of the Selection, Administration, Interpretation, and Reporting of Unidimensional Pain Intensity Scales. *Pain Res Manag*. 2017; **2017**: 7603758.
19. Ostojic K, Paget SP, Morrow AM. Management of pain in children and adolescents with cerebral palsy: a systematic review. *Developmental Medicine And Child Neurology*. 2019; **61**(3): 315-21.
20. Liossi C, Howard RF. Pediatric Chronic Pain: Biopsychosocial Assessment and Formulation. *Pediatrics*. 2016; **138**(5): e20160331.
21. Harvey AR, McKinnon CT, Smith N, Ostojic K, Paget SP, Smith S, et al. Establishing consensus for the assessment of chronic pain in children and young people with cerebral palsy: a Delphi study. *Disabil Rehabil*. 2021; **44**(23): 7161–6.
22. Smith N. Chronic pain assessment for young people with cerebral palsy: Moving beyond pain intensity. *Dev Med Child Neurol*. 2023; **66**: 554–5.
23. Kingsnorth S, Orava T, Provvidenza C, Adler E, Ami N, Gresley-Jones T, et al. Chronic Pain Assessment Tools for Cerebral Palsy: A Systematic Review. *Pediatrics*. 2015; **136**(4): e947-60.
24. Smith NL, Smith MG, Gibson N, Imms C, Thornton Al, Harvey AR. Pain coping tools for children and young adults with a neurodevelopmental disability: A systematic review of measurement properties. *Dev Med Child Neurol*. 2023; **65**(3): 318-28.
25. Smith NL, Smith MG, Gibson N, Imms C, Thornton Al, Harvey AR. Pain coping tools for children and young adults with a neurodevelopmental disability: A systematic review of measurement properties. *Developmental Medicine & Child Neurology*. 2023; **65**(3): 318-28.
26. Riquelme I, Montoya P. Factors Affecting the Use of Pain-Coping Strategies in Individuals with Cerebral Palsy and Individuals with Typical Development. *Children*. 2023; **10**(1): 131.
27. Prinsen CAC, Mokkink LB, Bouter LM, Alonso J, Patrick DL, de Vet HCW, Terwee CB. COSMIN guideline for systematic reviews of patient-reported outcome measures. *Qual Life Res*. 2018; **27**(5): 1147-57.
28. Mokkink LB, Boers M, van der Vleuten CPM, Bouter LM, Alonso J, Patrick DL, et al. COSMIN Risk of Bias tool to assess the quality of studies on reliability or measurement error of outcome measurement instruments: a Delphi study. *BMC Med Res Methodol*. 2020; **20**(1): 293.
29. Mokkink LB, de Vet HCW, Prinsen CAC, Patrick DL, Alonso J, Bouter LM, Terwee CB. COSMIN Risk of Bias checklist for systematic reviews of Patient-Reported Outcome Measures. *Quality of Life Research*. 2018; **27**(5): 1171-9.
30. Terwee CB, Prinsen CAC, Chiarotto A, Westerman MJ, Patrick DL, Alonso J, et al. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Qual Life Res*. 2018; **27**(5): 1159-70.
31. de Vet HCW, Terwee CB, Mokkink LB, Knol DL. Measurement in Medicine: A Practical Guide. Cambridge: Cambridge University Press; 2011.
32. Emerson ND, Bursch B. Communicating with Youth about Pain: Developmental Considerations. *Children (Basel)*. 2020; **7**(10): 1.

33. Letzkus L, Fehlings D, Ayala L, Byrne R, Gehred A, Maitre NL, et al. A Systematic Review of Assessments and Interventions for Chronic Pain in Young Children With or at High Risk for Cerebral Palsy. *Journal of Child Neurology*. 2021: 883073821996916-.
34. Sultan T, Wong C. Presence and grade of undertreatment of pain in children with cerebral palsy. *Scand J Pain*. 2023; **23**(3): 546-52.
35. Mutch L, Alberman E, Hagberg B, Kodama K, Perat MV. Cerebral Palsy Epidemiology: Where are We Now and Where are We Going? *Developmental Medicine And Child Neurology*. 1992; **34**(6): 547-51.
36. Australian Cerebral Palsy Register. Report of the Australian Cerebral Palsy Register: Birth years 1995-2016. January 2023.
37. Hinwood GL, Loftus H, Shepherd DA, Guzys A, Reddihough DS, Reid SM. Survival of individuals with cerebral palsy in Victoria, Australia: A longitudinal cohort study spanning four decades. *Developmental Medicine & Child Neurology*. 2023; **65**(4): 580-7.
38. Hutton JL, Pharoah PO. Life expectancy in severe cerebral palsy. *Arch Dis Child*. 2006; **91**(3): 254-8.
39. Colver A. Outcomes for people with cerebral palsy: life expectancy and quality of life. *Paediatrics and child health*. 2016; **26**(9): 383-6.
40. Ryan JM, Peterson MD, Ryan N, Smith KJ, O'connell NE, Liverani S, et al. Mortality due to cardiovascular disease, respiratory disease, and cancer in adults with cerebral palsy. *Developmental Medicine & Child Neurology*. 2019; **61**(8): 924-8.
41. McKinnon CT, Morgan PE, Antolovich GC, Clancy CH, Fahey MC, Harvey AR. Pain in children with dyskinetic and mixed dyskinetic/spastic cerebral palsy. *Developmental Medicine And Child Neurology*. 2020; **62**(11): 1294-301.
42. van Gorp M, Dallmeijer AJ, van Wely L, de Groot V, Terwee CB, Flens G, et al. Pain, fatigue, depressive symptoms and sleep disturbance in young adults with cerebral palsy. *Disability And Rehabilitation*. 2021; **43**(15): 2164-71.
43. Saleh MN, Almasri NA, Al Bakri B. Pain in children with cerebral palsy as reported by parents. *Child: Care, Health and Development*. 2023; **n/a**(n/a).
44. Rosenbaum PL, Walter SD, Hanna SE, Palisano RJ, Russell DJ, Raina P, et al. Prognosis for Gross Motor Function in Cerebral Palsy: Creation of Motor Development Curves. *JAMA : the journal of the American Medical Association*. 2002; **288**(11): 1357-63.
45. Smits DW, Gorter JW, Hanna SE, Dallmeijer AJ, van Eck M, Roebroeck ME, et al. Longitudinal development of gross motor function among Dutch children and young adults with cerebral palsy: an investigation of motor growth curves. *Developmental Medicine And Child Neurology*. 2013; **55**(4): 378-84.
46. Hanna SE, Rosenbaum PL, Bartlett DJ, Palisano RJ, Walter SD, Avery L, Russell DJ. Stability and decline in gross motor function among children and youth with cerebral palsy aged 2 to 21 years. *Developmental Medicine & Child Neurology*. 2009; **51**(4): 295-302.
47. Damiano DL. Activity, Activity, Activity: Rethinking Our Physical Therapy Approach to Cerebral Palsy. *Physical therapy*. 2006; **86**(11): 1534-40.
48. Sanger TD, Delgado MR, Gaebler-Spira D, Hallett M, Mink JW. Classification and Definition of Disorders Causing Hypertonia in Childhood. *Pediatrics (Evanston)*. 2003; **111**(1): e89-e97.
49. Rethlefsen SAPT, Ryan DDMD, Kay RMMD. Classification Systems in Cerebral Palsy. *Orthop Clin North Am*. 2010; **41**(4): 457-67.
50. Platt MJ, Krageloh-Mann I, Cans C. Surveillance of cerebral palsy in europe: reference and training manual. *Med Educ*. 2009; **43**(5): 495-6.

51. Report of the Australian Cerebral Palsy Register Birth Years 1995-2012. 2018 November 2018.
52. Surveillance of cerebral palsy in Europe: a collaboration of cerebral palsy surveys and registers. Surveillance of Cerebral Palsy in Europe (SCPE). *Dev Med Child Neurol*. 2000; **42**(12): 816-24.
53. Rice J, Skuza P, Baker F, Russo R, Fehlings D. Identification and measurement of dystonia in cerebral palsy. *Developmental Medicine And Child Neurology*. 2017; **59**(12): 1249-55.
54. Piscitelli D, Ferrarello F, Ugolini A, Verola S, Pellicciari L. Measurement properties of the Gross Motor Function Classification System, Gross Motor Function Classification System-Expanded & Revised, Manual Ability Classification System, and Communication Function Classification System in cerebral palsy: a systematic review with meta-analysis. *Developmental Medicine And Child Neurology*. 2021; **63**(11): 1251-61.
55. Pennington L, Virella D, Mjøen T, da Graça Andrada M, Murray J, Colver A, et al. Development of The Viking Speech Scale to classify the speech of children with cerebral palsy. *Research In Developmental Disabilities*. 2013; **34**(10): 3202-10.
56. Baranello G, Signorini S, Tinelli F, Guzzetta A, Pagliano E, Rossi A, et al. Visual Function Classification System for children with cerebral palsy: development and validation. *Developmental Medicine And Child Neurology*. 2020; **62**(1): 104-10.
57. Novak I, Hines M, Goldsmith S, Barclay R. Clinical prognostic messages from a systematic review on cerebral palsy. *Pediatrics*. 2012; **130**(5): e1285-e312.
58. Delacy MJ, Reid SM. Profile of associated impairments at age 5 years in Australia by cerebral palsy subtype and Gross Motor Function Classification System level for birth years 1996 to 2005. *Developmental Medicine And Child Neurology*. 2016; **58**: 50-6.
59. Raja SN, Carr DB, Cohen M, Finnerup NB, Flor H, Gibson S, et al. The revised International Association for the Study of Pain definition of pain: concepts, challenges, and compromises. *PAIN*. 2020; **161**(9).
60. Carr DB, Goudas LC. Acute pain. *Lancet*. 1999; **353**(9169): 2051-8.
61. Treede R-D, Rief W, Barke A, Aziz Q, Bennett MI, Benoliel R, et al. A classification of chronic pain for ICD-11. *PAIN*. 2015; **156**(6).
62. Chambers CT, Dol J, Tutelman PR, Langley CL, Parker JA, Cormier BT, et al. The prevalence of chronic pain in children and adolescents: a systematic review update and meta-analysis. *PAIN*. 2024.
63. Huguet A, Miró J. The Severity of Chronic Pediatric Pain: An Epidemiological Study. *The Journal of Pain*. 2008; **9**(3): 226-36.
64. Chin EM, Lenz C, Ye X, Campbell CM, Stashinko E, Jantzie LL, et al. Clinical Factors Associated With Chronic Pain in Communicative Adults With Cerebral Palsy: A Cross-Sectional Study. *Frontiers in pain research (Lausanne, Switzerland)*. 2020; **1**: 553026-.
65. Jarlman E, Hägglund G, Alriksson-Schmidt AI. Foot and lower leg pain in children and adults with cerebral palsy: a population-based register study on 5,122 individuals. *BMC Musculoskelet Disord*. 2024; **25**(1): 391.
66. Wong C. The Relationship between Pain and Spasticity and Tell-Tale Signs of Pain in Children with Cerebral Palsy. *Toxins (Basel)*. 2023; **15**(2).
67. Casey J, Rosenblad A, Rodby-Bousquet E. Postural asymmetries, pain, and ability to change position of children with cerebral palsy in sitting and supine: a cross-sectional study. *Disability And Rehabilitation*. 2020: 1-9.

68. Heinen F, Bonfert M, Kaňovský P, Schroeder AS, Chambers HG, Dabrowski E, et al. Spasticity-related pain in children/adolescents with cerebral palsy. Part 1: Prevalence and clinical characteristics from a pooled analysis. *Journal of Pediatric Rehabilitation Medicine*. 2022; **15**(1): 129-43.
69. Karkenny AJ, Mackey C, Sharkey MS. What Do We Really Know About the Natural History of Spastic Hip Dysplasia and Pain in Total-involvement Cerebral Palsy? A Systematic Review. *Journal of Pediatric Orthopaedics*. 9900.
70. Blackman JA, Svensson CI, Marchand S. Pathophysiology of chronic pain in cerebral palsy: implications for pharmacological treatment and research. *Developmental Medicine And Child Neurology*. 2018; **60**(9): 861-5.
71. Shekari Z, Sadeghian Afarani R, Fatorehchy S, Bakhshi E, Shahshahani S, Mousavi E. Relationship Between Postural Asymmetry, Balance, and Pain in Children With Spastic Cerebral Palsy. *Pediatr Neurol*. 2024; **155**: 84-90.
72. Hanaoka C, Gaebler-Spira D, Pichika R, Jayabalan P. Comparative study of the pain, function and biomarkers of joint disease in the transition to adulthood in individuals with and without cerebral palsy. *American Journal of Physical Medicine & Rehabilitation*. 2024; **103**(2): 110-6.
73. Ostojic K, Sharp NL, Paget SP, Morrow AM. Lived experiences of pain in children and young people with cerebral palsy. *Developmental Medicine And Child Neurology*. 2022; **64**(3): 364-71.
74. Dubois A, Hall C, Courtois-Communier E, Brasseur A, Cacioppo M, Brochard S. Incidence and risk factors for care-related pain in children with physical disabilities. *Eur J Phys Rehabil Med*. 2023; **59**(3): 414-24.
75. Peterson MD, Haapala HJ, Kratz A. Pain Phenotypes in Adults Living With Cerebral Palsy. *Neurol Clin Pract*. 2021; **11**(6): e848-e55.
76. Vinkel MN, Rackauskaite G, Finnerup NB. Classification of pain in children with cerebral palsy. *Dev Med Child Neurol*. 2021.
77. Larsen SM, Terjesen T, Jahnsen RB, Diseth TH, Ramstad K. Health-related quality of life in adolescents with cerebral palsy; a cross-sectional and longitudinal population-based study. *Child : care, health & development*. 2022.
78. Shearer HM, Côté P, Hogg-Johnson S, Fehlings DL. A good night's sleep: pain trajectories and sleep disturbance in children with cerebral palsy. *Journal of clinical sleep medicine*. 2024.
79. Carozza L, Anderson-Mackay E, Blackmore AM, Kirkman HA, Ou J, Smith N, Love S. Chronic Pain in Young People With Cerebral Palsy: Activity Limitations and Coping Strategies. *Pediatric Physical Therapy*. 2022.
80. Sarmiento CA, Gannotti ME, Gross PH, Thorpe DE, Hurvitz EA, Noritz GH, et al. Adults with cerebral palsy and chronic pain experience: A cross-sectional analysis of patient-reported outcomes from a novel North American registry. *Disability and Health Journal*. 2023: 101546.
81. Abid M, Cherni Y, Batcho CS, Traverse E, Lavoie MD, Mercier C. Facilitators and barriers to participation in physical activities in children and adolescents living with cerebral palsy: a scoping review. *Disability And Rehabilitation*. 2023; **45**(25): 4322-37.
82. Van Der Slot WMA, Nieuwenhuijsen C, Van Den Berg-Emons RJG, Bergen MP, Hilberink SR, Stam HJ, Roebroeck ME. Chronic pain, fatigue, and depressive symptoms in adults with spastic bilateral cerebral palsy. *Developmental Medicine & Child Neurology*. 2012; **54**(9): 836-42.
83. Sattoe JNT, Hilberink SR. Impairments and comorbidities in adults with cerebral palsy and spina bifida: a meta-analysis. *Front Neurol*. 2023; **14**.

84. Brian GB, Sonali S, Neil C, Janice M, Pip L, Richard L, Anthony JA. The impact of ageing on adults with cerebral palsy: the results of a national online survey. *BJGP Open*. 2023; **7**(4): BJGPO.2023.0028.
85. Shearer HM, Verville L, Côté P, Hogg-Johnson S, Fehlings DL. Clinical course of pain intensity in individuals with cerebral palsy: A prognostic systematic review. *Developmental Medicine And Child Neurology*. 2022.
86. Bell B, Shah S, Coulson N, McLaughlin J, Logan P, Luke R, Avery AJ. The impact of ageing on adults with cerebral palsy. *BJGP Open*. 2023.
87. Christensen R, MacIntosh A, Switzer L, Fehlings D. Change in pain status in children with cerebral palsy. *Developmental Medicine And Child Neurology*. 2017; **59**(4): 374-9.
88. Larsen SM, Terjesen T, Jahnsen RB, Ramstad K. Recurrent pain in adolescents with cerebral palsy: a longitudinal population-based study. *Developmental Medicine And Child Neurology*. 2022; **64**(3): 357-63.
89. Rochani HD, Modlesky CM, Li L, Weissman B, Vova J, Colquitt G. Association of Chronic Pain with Participation in Motor Skill Activities in Children with Cerebral Palsy. *JAMA Netw Open*. 2021; **4**(7): e2115970-e.
90. Lennon N, Kalisperis F, Church C, Niiler T, Miller F, Biermann I, et al. Self-reported Health-related Quality of Life in Adolescents With Cerebral Palsy. *Journal of Pediatric Orthopaedics*. 2023.
91. Engel JM, Jensen MP, Hoffman AJ, Kartin D. Pain in persons with cerebral palsy: extension and cross validation. *Archives Of Physical Medicine And Rehabilitation*. 2003; **84**(8): 1125-8.
92. Schmidt S, Markwart H, Rapp M, Guyard A, Arnaud C, Fauconnier J, et al. Quality of life and mental health in emerging adults with cerebral palsy compared to the general population. *Health and Quality of Life Outcomes*. 2022; **20**(1): 61.
93. Testani D, McMorris CA, Clark CA, Sanguino H, Condliffe EG, Noel ME, et al. Investigating physiological symptoms associated with mental health symptoms in youth with cerebral palsy: An observational study. *Research In Developmental Disabilities*. 2024; **151**: 104783.
94. Asuman D, Gerdtham U-G, Alriksson-Schmidt AI, Rodby-Bousquet E, Andersen GL, Jarl J. Pain and labor outcomes: A longitudinal study of adults with cerebral palsy in Sweden. *Disability and Health Journal*. 2023; **16**(3): 101479.
95. Shih STF, Tonmukayakul U, Imms C, Reddihough D, Graham HK, Cox L, Carter R. Economic evaluation and cost of interventions for cerebral palsy: a systematic review. *Developmental Medicine & Child Neurology*. 2018; **60**(6): 543-58.
96. Nous Group. Value of Physiotherapy in Australia. Australian Physiotherapy Association; 2020.
97. McGrath PJ, Walco GA, Turk DC, Dworkin RH, Brown MT, Davidson K, et al. Core Outcome Domains and Measures for Pediatric Acute and Chronic/Recurrent Pain Clinical Trials: PedIMMPACT Recommendations. *J Pain*. 2008; **9**(9): 771-83.
98. Palermo TM, Walco GA, Paladhi UR, Birnie KA, Crombez G, de la Vega R, et al. Core outcome set for pediatric chronic pain clinical trials: results from a Delphi poll and consensus meeting. *PAIN*. 2021; **162**(10).
99. Trottier ED, Ali S, Doré-Bergeron M-J, Chauvin-Kimoff L. Best practices in pain assessment and management for children. *Paediatrics & Child Health*. 2022; **27**(7): 429-37.
100. Greenough MJ, Jibb L, Lewis KB, Bucknall T, Lamontagne C, Demery Varin M, et al. A systematic review of the biopsychosocial dimensions affected by chronic pain in

children and adolescents: identifying reliable and valid pediatric multidimensional chronic pain assessment tools. *Pain Rep*. 2023; **8**(6): e1099.
101. McKinnon C, White J, Harvey A, Antolovich G, Morgan P. Caregiver perspectives of managing chronic pain in children and adolescents with dyskinetic and mixed dyskinetic/spastic CP with communication limitations. *Journal of Pediatric Rehabilitation Medicine*. 2021: 1-13.
102. McKinnon CT, White JH, Morgan PE, Antolovich GC, Clancy CH, Fahey MC, Harvey AR. The lived experience of chronic pain and dyskinesia in children and adolescents with cerebral palsy. *BMC Pediatrics*. 2020; **20**(1): 125-.
103. Greenberg A. Pain and cerebral palsy: the need for a broader perspective to intervention. *Developmental Medicine And Child Neurology*. 2022; **64**(3): 283-.
104. Gross PH, Bailes AF, Horn SD, Hurvitz EA, Kean J, Shusterman M. Setting a patient-centered research agenda for cerebral palsy: a participatory action research initiative. *Developmental Medicine And Child Neurology*. 2018; **60**(12): 1278-84.
105. Almoajil H, Toye F, Dawes H, Pierce J, Meaney A, Baklouti A, et al. Outcomes of importance to children and young adults with cerebral palsy, their parents and health professionals following lower limb orthopaedic surgery: A qualitative study to inform a Core Outcome Set. *Health Expectations*. 2022; **25**(3): 925-35.
106. Schiariti V, Oberlander TF. Evaluating pain in cerebral palsy: comparing assessment tools using the International Classification of Functioning, Disability and Health. *Disability And Rehabilitation*. 2019; **41**(22): 2622-9.
107. Stevens B. Revisions to the IASP definition of pain—What does this mean for children? *Paediatric and neonatal pain*. 2021; **3**(3): 101-5.
108. Williams E, Coghill J. Understanding and managing pain in children with cerebral palsy. *Paediatrics and child health*. 2022.
109. World Health Organisation. International classification of functioning, disability and health children and youth version : ICF-CY. Geneva, Switzerland: World Health Organization; 2007.
110. Dan B. The ICF as a socio-psycho-biological model for the full participation of disabled individuals. *Developmental Medicine & Child Neurology*. **n/a**(n/a).
111. Heydenrijk-Kikkert MA, Schmidt AKK, Pangalila R, De Wit M-CY, van Haren NEM, Van Veelen M-LC, Roebroeck ME. Meaningful outcomes for children and their caregivers attending a paediatric brain centre. *Developmental Medicine And Child Neurology*. 2023.
112. Noyek S, Jessa JS, Faulkner V, Boerner KE, Dewan T, Doyle D, et al. A systematic review of self and observer assessment of pain and related functioning in youth with brain-based developmental disabilities. *PAIN*. 2024; **165**(3): 523-36.
113. Smith NL, Gibson N, Bear N, Thornton AL, Imms C, Smith MG, Harvey AR. Measurement properties and feasibility of chronic pain assessment tools for use with children and young people with cerebral palsy. *Disability And Rehabilitation*. 2024: 1-15.
114. Caravau H, Rosa AF, Rocha NP, Silva AG. Pain assessment in cerebral palsy: a systematic review of measurement properties and evaluation using the COSMIN checklist. *Disability And Rehabilitation*. 2020: 1-11.
115. Schiariti V, Shierk A, Stashinko EE, Sukal-Moulton T, Feldman RS, Aman C, et al. Cerebral palsy pain instruments: Recommended tools for clinical research studies by the National Institute of Neurological Disorders and Stroke Cerebral Palsy Common Data Elements project. *Developmental Medicine & Child Neurology*. 2024; **66**: 610-22.

116. Gannotti ME, Law M, Bailes AF, O'Neil ME, Williams U, Direzze B. Comparative Effectiveness Research and Children with Cerebral Palsy: Identifying a Conceptual Framework and Specifying Measures. *Pediatric Physical Therapy*. 2016; **28**(1): 58-69.
117. Organization HS. National Standard of Canada: Pediatric Pain Management. 2023.
118. Barney CC, Andersen RD, Defrin R, Genik LM, McGuire BE, Symons FJ. Challenges in pain assessment and management among individuals with intellectual and developmental disabilities. *Pain reports*. 2020; **5**(4): e821.
119. Breau LM, MacLaren J, McGrath PJ, Camfield CS, Finley GA. Caregivers' beliefs regarding pain in children with cognitive impairment: relation between pain sensation and reaction increases with severity of impairment. *Clin J Pain*. 2003; **19**(6): 335-44.
120. Özcan F, Ünsal Delialioğlu S, Özel S, Demir Y. Perception of pain in patients with adolescent cerebral palsy: self report or parent's report. *Somatosens Mot Res*. 2021: 1-6.
121. Davis E, Mackinnon A, Waters E. Parent proxy-reported quality of life for children with cerebral palsy: is it related to parental psychosocial distress? *Child : care, health & development*. 2012; **38**(4): 553-60.
122. Noyek S, Vowles C, Batorowicz B, Davies C, Fayed N. Direct assessment of emotional well-being from children with severe motor and communication impairment: a systematic review. *Disabil Rehabil Assist Technol*. 2022; **17**(5): 501-14.
123. Guard A, Konz M, Smith RO, Engel JM, Keating T. The development of an iPad application for the pain assessment of individuals with developmental disabilities and complex communication needs. *Assistive technology*. 2021; **33**(4): 178-89.
124. Holmes MM, Lewith G, Newell D, Field J, Bishop FL. The impact of patient-reported outcome measures in clinical practice for pain: a systematic review. *Qual Life Res*. 2017; **26**(2): 245-57.
125. Dahlhamer J, Lucas J, Zelaya C, Nahin R, Mackey S, DeBar L, et al. Prevalence of Chronic Pain and High-Impact Chronic Pain Among Adults — United States, 2016. *MMWR Morb Mortal Wkly Rep*. 2018; **67**(36): 1001-6.
126. King S, Chambers CT, Huguet A, MacNevin RC, McGrath PJ, Parker L, MacDonald AJ. The epidemiology of chronic pain in children and adolescents revisited: a systematic review. *PAIN*. 2011; **152**(12): 2729-38.
127. Murray CB, de la Vega R, Murphy LK, Kashikar-Zuck S, Palermo TM. The prevalence of chronic pain in young adults: a systematic review and meta-analysis. *PAIN*. 2021; **163**(9): e972–e84.
128. van der Slot WMA, Benner JL, Brunton L, Engel JM, Gallien P, Hilberink SR, et al. Pain in adults with cerebral palsy: A systematic review and meta-analysis of individual participant data. *Ann Phys Rehabil Med*. 2021; **64**(3): 101359.
129. Atee M, Morris T, Macfarlane S, Cunningham C. Prevalence of Pain in Dementia Subtypes: Data from a National Sample of Aged Care Residents in Australia. *International Psychogeriatrics*. 2021; **33**(S1): 43.
130. Kroenke K, Krebs EE, Turk D, Von Korff M, Bair MJ, Allen KD, et al. Core Outcome Measures for Chronic Musculoskeletal Pain Research: Recommendations from a Veterans Health Administration Work Group. *Pain Med*. 2019; **20**(8): 1500-8.
131. Nieuwenhuijsen C, van Der Laar Y, Donkervoort M, Nieuwstraten W, Roebroeck ME, Stam HJ. Unmet needs and health care utilization in young adults with cerebral palsy. *Disability And Rehabilitation*. 2008; **30**(17): 1254.

132. Karayannis NV, Sturgeon JA, Chih-Kao M, Cooley C, Mackey SC. Pain interference and physical function demonstrate poor longitudinal association in people living with pain: a PROMIS investigation. *PAIN*. 2017; **158**(6): 1063-8.
133. Wilson IB, Cleary PD. Linking Clinical Variables With Health-Related Quality of Life: A Conceptual Model of Patient Outcomes. *JAMA : the journal of the American Medical Association*. 1995; **273**(1): 59-65.
134. Andersen RD, Langius-Eklöf A, Nakstad B, Bernklev T, Jylli L. The measurement properties of pediatric observational pain scales: A systematic review of reviews. *International journal of nursing studies*. 2017; **73**: 93-101.
135. Stolee P, Hillier LM, Esbaugh J, Bol N, McKellar L, Gauthier N. Instruments for the assessment of pain in older persons with cognitive impairment. *Journal of the American Geriatrics Society*. 2005; **53**(2): 319-26.
136. van Herk R, van Dijk M, Baar FPM, Tibboel D, de Wit R. Observation scales for pain assessment in older adults with cognitive impairments or communication difficulties. *Nurs Res*. 2007; **56**(1): 34-43.
137. Voepel-Lewis T, Malviya S, Tait AR, Merkel S, Foster R, Krane EJ, Davis PJ. A comparison of the clinical utility of pain assessment tools for children with cognitive impairment. *Anesth Analg*. 2008; **106**(1): 72.
138. Aubin M, Giguere A, Hadjistavropoulos T, Verreault R. The systematic evaluation of instruments designed to assess pain in persons with limited ability to communicate. *L'evaluation systematique des instruments pour mesurer la douleur chez les personnes agees ayant des capacites reduites a communiquer*. 2007; **12**(3): 195-203.
139. Coca SM, Abeldano Zuniga RA. Instruments for pain assessment in patients with advanced dementia: A systematic review of the evidence for Latin America. *Palliative & supportive care*. 2020; **18**(6): 741-7.
140. Herr K, Bjoro K, Decker S. Tools for assessment of pain in nonverbal older adults with dementia: a state-of-the-science review. *Journal of Pain and Symptom Management*. 2006; **31**(2): 170-92.
141. Storti M, Bertozzo E, Dal Santo P, Mari M, Dal Cengio L, Forni C, et al. [The measurement of pain in patients with dementia: a review of the most appropriate tools]. *La misurazione del dolore nel paziente affetto da demenza: una rassegna degli strumenti piu appropriati*. 2014; **105**(4): 166-74.
142. Zwakhalen SMG, Hamers JPH, Abu-Saad HH, Berger MPF. Pain in elderly people with severe dementia: a systematic review of behavioural pain assessment tools. *BMC Geriatr*. 2006; **6**: 3.
143. Smith TO, Harvey K. Psychometric properties of pain measurements for people living with dementia: a COSMIN systematic review. *European geriatric medicine*. 2022; **13**: 1029-45.
144. Morasco B, Lovejoy T, Hyde S, Shull S, Dobscha S. Limitations of pain numeric rating scale scores collected during usual care: Need for enhanced assessment. *Journal of Pain*. 2018; **19**(3 Supplement 1): S33.
145. von Baeyer CL, Spagrud LJ. Systematic review of observational (behavioral) measures of pain for children and adolescents aged 3 to 18 years. *PAIN*. 2007; **127**(1-2): 140-50.
146. Page MJ, McKenzie JE, Bossuyt PM, Boutron I, Hoffmann TC, Mulrow CD, et al. The PRISMA 2020 statement: an updated guideline for reporting systematic reviews. *BMJ*. 2021; **372**: n71.

147. Terwee CB, Jansma EP, Riphagen II, de Vet HCW. Development of a methodological PubMed search filter for finding studies on measurement properties of measurement instruments. *Quality of Life Research*. 2009; **18**(8): 1115-23.
148. Eccleston C, McCracken LM, Jordan A, Sleed M. Development and preliminary psychometric evaluation of the parent report version of the Bath Adolescent Pain Questionnaire (BAPQ-P): A multidimensional parent report instrument to assess the impact of chronic pain on adolescents. *Pain (Amsterdam)*. 2007; **131**(1): 48-56.
149. Greig ML, Chow E, Bovett G, Abdolell M, Gillies C. Level of Concordance Between Proxy and Cancer Patient Ratings in Brief PainInventory. *Supportive Cancer Therapy*. 2005; **3**(1): 54-8.
150. Palermo TM, Witherspoon D, Valenzuela D, Drotar DD. Development and validation of the Child Activity Limitations Interview: A measure of pain-related functional impairment in school-age children and adolescents. *PAIN*. 2004; **109**(3): 461-70.
151. Palermo TM, Lewandowski AS, Long AC, Burant CJ. Validation of a self-report questionnaire version of the Child Activity Limitations Interview (CALI): The CALI-21. *PAIN*. 2008; **139**(3): 644-52.
152. Hainsworth KR, Davies WH, Khan KA, Weisman SJ. Development and preliminary validation of the child activity limitations questionnaire: flexible and efficient assessment of pain-related functional disability. *The journal of pain : official journal of the American Pain Society*. 2007; **8**(9): 746-52.
153. Pickering G, Gibson SJ, Serbouti S, Odetti P, Ferraz Goncalves J, Gambassi G, et al. Reliability study in five languages of the translation of the pain behavioural scale Doloplus. *Eur J Pain*. 2010; **14**(5): 545.e1-10.
154. Barney CC, Stibb SM, Merbler AM, Summers RLS, Deshpande S, Krach LE, Symons FJ. Psychometric properties of the brief pain inventory modified for proxy report of pain interference in children with cerebral palsy with and without cognitive impairment. *Pain Rep*. 2018; **3**(4): e666.
155. Hunt A, Wisbeach A, Seers K, Goldman A, Crichton N, Perry L, Mastroyannopoulou K. Development of the Paediatric Pain Profile: Role of Video Analysis and Saliva Cortisol in Validating a Tool to Assess Pain in Children with Severe Neurological Disability. *Journal of Pain and Symptom Management*. 2007; **33(3)**: 276-89.
156. Santos M, Santanelli JP, Zempsky WT. Preliminary Validation of the Pain Burden Inventory in a Pediatric Chronic Pain Population. *Clin J Pain*. 2021; **37**(11): 820-4.
157. Martin S, Nelson Schmitt S, Wolters PL, Abel B, Toledo-Tamula MA, Baldwin A, et al. Development and validation of the English Pain Interference Index and Pain Interference Index-Parent report. *Pain Med*. 2015; **16**(2): 367-73.
158. Irwin DE, Gross HE, Stucky BD, Thissen D, DeWitt EM, Lai JS, et al. Development of six PROMIS pediatrics proxy-report item banks. *Health and Quality of Life Outcomes*. 2012; **10**: 22.
159. Amtmann D, Cook KF, Jensen MP, Chen W-H, Choi S, Revicki D, et al. Development of a PROMIS item bank to measure pain interference. *Pain (Amsterdam)*. 2010; **150**(1): 173-82.
160. Holley AL, Zhou C, Wilson AC, Hainsworth K, Palermo TM. The CALI-9: A brief measure for assessing activity limitations in children and adolescents with chronic pain. *PAIN*. 2018; **159**(1): 48-56.
161. Ando C, Hishinuma M. Development of the Japanese DOLOPLUS-2: a pain assessment scale for the elderly with Alzheimer's disease. *Psychogeriatrics*. 2010; **10**(3): 131-7.

162. Chen YH, Lin LC, Watson R. Evaluation of the psychometric properties and the clinical feasibility of a Chinese version of the Doloplus-2 scale among cognitively impaired older people with communication difficulty. *International journal of nursing studies*. 2010; **47**(1): 78-88.
163. Chen YH, Lin LC, Watson R. Validating nurses' and nursing assistants' report of assessing pain in older people with dementia. *Journal of Clinical Nursing*. 2010; **19**(1-2): 42-52.
164. Holen JC, Saltvedt I, Fayers PM, Bjornnes M, Stenseth G, Hval B, et al. The Norwegian Doloplus-2, a tool for behavioural pain assessment: translation and pilot-validation in nursing home patients with cognitive impairment. *Palliat Med*. 2005; **19**(5): 411-7.
165. Holen JC, Saltvedt I, Fayers PM, Hjermstad MJ, Loge JH, Kaasa S. Doloplus-2, a valid tool for behavioural pain assessment? *BMC Geriatr*. 2007; **7**: 29.
166. Torvik K, Kaasa S, Kirkevold O, Saltvedt I, Holen JC, Fayers P, Rustoen T. Validation of Doloplus-2 among nonverbal nursing home patients--an evaluation of Doloplus-2 in a clinical setting. *BMC Geriatr*. 2010; **10**: 9.
167. Lepetit-Vitou V, Gely-Nargeot M-C, Mimoun N, Million E, Gayral A, Bayard S. Pain assessment of nursing home's residents by certified nursing assistants: A real-world study. *Evaluation de la douleur des residents en Ehpad par des aides-soignantes: Une etude en condition ecologique*. 2019; **17**(4): 423-8.
168. Pautex S, Herrmann FR, Michon A, Giannakopoulos P, Gold G. Psychometric properties of the Doloplus-2 observational pain assessment scale and comparison to self-assessment in hospitalized elderly. *Clin J Pain*. 2007; **23**(9): 774-9.
169. Likar R, Pipam W, Neuwersch S, Kostenberger M, Pinter G, Gatternig C, Marksteiner J. [Pain measurement in cognitively impaired patients with the Doloshort scale]. *Schmerzmessung bei kognitiv beeintrachtigten Patienten mit der Doloshort-Skala*. 2015; **29**(4): 440-4.
170. Zare M, Tagharrobi Z, Sharifi K, Sooki Z, Abolhasani J. Psychometric evaluation of the Persian version of the Doloplus-2 (P-Doloplus-2) scale in elderly with dementia. *Turk J Med Sci*. 2020; **50**(4): 953-62.
171. Zwakhalen SMG, Hamers JPH, Berger MPF. The psychometric quality and clinical usefulness of three pain assessment tools for elderly people with dementia. *PAIN*. 2006; **126**(1-3): 210-20.
172. Pasin S, Avila F, de Cavata T, Hunt A, Heldt E. Cross-cultural translation and adaptation to Brazilian Portuguese of the paediatric pain profile in children with severe cerebral palsy. *Journal of Pain and Symptom Management*. 2013; **45**(1): 120-8.
173. Vargus-Adams JN, Farrell J, Morgan-Dewitt E, Jacobson C. 'I don't think she sees herself the way she truly is': A qualitative analysis of concordance/ discordance in child and parent representations and reports of physical functioning associated with cerebral palsy. *Developmental Medicine & Child Neurology*. 2012; **54**(SUPPL. 6): 51.
174. Vargus-Adams JN, Jacobson C, Carle AC, Farrell J, Morgan-Dewitt E. Validation of PROMIS in childhood cerebral palsy. *Developmental Medicine & Child Neurology*. 2013; **55**(SUPPL. 3): 40-1.
175. Amtmann D, Bamer AM, McMullen K, Stewart BT, Kazis LE, Rencken CA. Agreement between proxy-and self-report PROMIS scores in pediatric burn survivors. *Journal of Burn Care and Research*. 2021; **42**(SUPPL 1): S65.
176. Bamer AM, McMullen K, Wolf SE, Stewart BT, Kazis L, Rencken CA, Amtmann D. Agreement between proxy- and self-report scores on PROMIS health-related quality of life domains in pediatric burn survivors: a National Institute on Disability, Independent

Living, and Rehabilitation Research Burn Model System Study. *Qual Life Res*. 2021; **30**(7): 2071-80.
177. Bertisch H, Rivara FP, Kisala PA, Wang J, Yeates KO, Durbin D, et al. Psychometric evaluation of the pediatric and parent-proxy Patient-Reported Outcomes Measurement Information System and the Neurology and Traumatic Brain Injury Quality of Life measurement item banks in pediatric traumatic brain injury. *Qual Life Res*. 2017; **26**(7): 1887-99.
178. Brandon TG, Becker BD, Bevans KB, Weiss PF. Patient-Reported Outcomes Measurement Information System Tools for Collecting Patient-Reported Outcomes in Children With Juvenile Arthritis. *Arthritis Care Res (Hoboken)*. 2017; **69**(3): 393-402.
179. Trachtman R, Murray ET, Szymonifka J, Adams AB, Pan N, Taber SF, et al. Do patient reported outcome measurement information system (PROMIS) computer adaptive tests correlate with disease activity in juvenile idiopathic arthritis? *Annals of the Rheumatic Diseases*. 2018; **77**: 856-7.
180. Patel R, Esparza V, Lai JS, Gray E, Chang R, Cella D, Ardalan K. Comparison of PROMIS computerized adaptive testingadministered item banks versus fixed short forms in Juvenile Myositis. *Quality of Life Research*. 2019; **29**(SUPPL 1): S22.
181. Wolfe M, Robinson A, Lai JS, Coles T, Gray E, Chang R, et al. Estimation of Clinically Important Diff erences in Patient-Reported Outcomes Measurement Information System (PROMIS) Measures in Juvenile Myositis. *Arthritis and Rheumatology*. 2020; **72**(SUPPL 10): 2278-81.
182. Lapin BR, Thompson NR, Schuster A, Honomichl R, Katzan IL. The validity of proxy responses on patient-reported outcome measures: Are proxies a reliable alternative to stroke patients' self-report? *Qual Life Res*. 2021; **30**(6): 1735-45.
183. Alvarez-Nebreda ML, Heng M, Rosner B, McTague M, Javedan H, Harris MB, Weaver MJ. Reliability of Proxy-reported Patient-reported Outcomes Measurement Information System Physical Function and Pain Interference Responses for Elderly Patients With Musculoskeletal Injury. *J Am Acad Orthop Surg*. 2019; **27**(4): e156-e65.
184. Scott EL, Foxen-Craft E, Caird M, Phillíben R, deSebour T, Currier E, Voepel-Lewis T. Parental Proxy PROMIS Pain Interference Scores are Only Modestly Concordant With Their Child's Scores: An Effect of Child Catastrophizing. *Clin J Pain*. 2020; **36**(1): 1-7.
185. Neville C, Ostini R. A psychometric evaluation of three pain rating scales for people with moderate to severe dementia. *Pain management nursing : official journal of the American Society of Pain Management Nurses*. 2014; **15**(4): 798-806.
186. Hunt A, Goldman A, Seers K, Crichton N, Mastroyannopoulou K, Moffat V, et al. Clinical validation of the paediatric pain profile. *Developmental Medicine & Child Neurology*. 2004; **46**: 9-18.
187. Cohen LL, Vowles KE, Eccleston C. Adolescent chronic pain-related functioning: Concordance and discordance of mother-proxy and self-report ratings. *European Journal of Pain*. 2010; **14**(8): 882-6.
188. Goddard JM, Robinson J, Hiscock R. Routine use of the Bath Adolescent Pain Questionnaire in a paediatric pain clinic. *British Journal of Pain*. 2021; **15**(2): 155-62.
189. Varni JW, Thissen D, Stucky BD, Liu Y, Magnus B, Quinn H, et al. PROMIS®Parent Proxy Report Scales for children ages 5–7 years: an item response theory analysis of differential item functioning across age groups. *Quality of Life Research*. 2014; **23**(1): 349-61.
190. Quinn H, Thissen D, Liu Y, Magnus B, Lai J-S, Amtmann D, et al. Using item response theory to enrich and expand the PROMIS® pediatric self report banks. *Health and Quality of Life Outcomes*. 2014; **12**(1): 160.

191. Birnie KA, Richardson PA, Rajagopalan AV, Bhandari RP. Factors Related to Agreement Between Child and Caregiver Report of Child Functioning With Chronic Pain: PROMIS Pediatric and Parent Proxy Report. *Clin J Pain*. 2020; **36**(3): 203-12.
192. Alcantara J, Ohm J, Alcantara J. Comparison of pediatric self reports and parent proxy reports utilizing PROMIS: Results from a chiropractic practice-based research network. *Complement Ther Clin Pract*. 2017; **29**: 48-52.
193. Hanania JW. Protective parental influences in pediatric chronic pain. *Dissertation Abstracts International: Section B: The Sciences and Engineering*. 2021; **82**(10-B).
194. Varni JW, Thissen D, Stucky BD, Liu Y, Magnus B, He J, et al. Item-level informant discrepancies between children and their parents on the PROMIS pediatric scales. *Quality of Life Research: An International Journal of Quality of Life Aspects of Treatment, Care & Rehabilitation*. 2015; **24**(8): 1921-37.
195. Engel JM, Kartin D, Carter GT, Jensen MP, Jaffe KM. Pain in youths with neuromuscular disease. *Am J Hosp Palliat Care*. 2009; **26**(5): 405-12.
196. Mokkink LB, Prinsen CA, Bouter LM, Vet HC, Terwee CB. The COnsensus-based Standards for the selection of health Measurement INstruments (COSMIN) and how to select an outcome measurement instrument. *Braz J Phys Ther*. 2016; **20**(2): 105-13.
197. Mokkink LB, Prinsen CA, Patrick DL, Alonso J, Bouter LM, de Vet HC, Terwee CB. COSMIN Study Design checklist for Patient-reported outcome measurement instruments. Amsterdam, The Netherlands2019.
198. Walker LS, Greene JW. The functional disability inventory: Measuring a neglected dimension of child health status. *Journal of Pediatric Psychology*. 1991; **16**(1): 39-58.
199. Üstün TB. Measuring health and disability manual for WHO Disability Assessment Schedule WHODAS 2.0. Geneva: World Health Organization; 2010.
200. Breau LM, McGrath PJ, Camfield CS, Finley GA. Psychometric properties of the non-communicating children's pain checklist-revised. *PAIN*. 2002; **99**(1-2): 349-57.
201. Skarsaune SN. Self-determination of people with profound intellectual and multiple disabilities. *Developmental Medicine & Child Neurology*. 2023; **65**(1): 16-23.
202. Smith MG, Farrar LC, Gibson RJ, Russo RN, Harvey AR. Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties. *Developmental Medicine & Child Neurology*. 2023.
203. Harvey AR, McKinnon CT, Smith N, Ostojic K, Paget SP, Smith S, et al. Establishing consensus for the assessment of chronic pain in children and young people with cerebral palsy: a Delphi study. *Disabil Rehabil*. 2021: 1-6.
204. Simons LE, Kaczynski KJ. The Fear Avoidance model of chronic pain: examination for pediatric application. *J Pain*. 2012; **13**(9): 827-35.
205. Fisher E, Heathcote LC, Eccleston C, Simons LE, Palermo TM. Assessment of Pain Anxiety, Pain Catastrophizing, and Fear of Pain in Children and Adolescents With Chronic Pain: A Systematic Review and Meta-Analysis. *J Pediatr Psychol*. 2018; **43**(3): 314-25.
206. Fisher E, Law E, Dudeney J, Palermo TM, Stewart G, Eccleston C. Psychological therapies for the management of chronic and recurrent pain in children and adolescents. *Cochrane Database Syst Rev*. 2018; **9**(9): Cd003968.
207. Jensen MP, Engel JM, Schwartz L. Coping with Cerebral Palsy Pain: A Preliminary Longitudinal Study. *Pain Med*. 2006; **7**(1): 30-7.
208. Dudgeon BJ, Tyler EJ, Rhodes LA, Jensen MP. Managing usual and unexpected pain with physical disability: a qualitative analysis. *The American Journal of Occupational Therapy*. 2006; **60**(1): 92-103.

209. Rinde ER, Anderzén-Carlsson A, Jahnsen RB, Andersen RD. "I have to obey my pain" - children's experiences of pain burden in cerebral palsy. *Disabil Rehabil*. 2023; **46**(6): 1112–20.
210. Smith MG, Farrar LC, Gibson RJ, Russo RN, Harvey AR. Chronic pain interference assessment tools for children and adults who are unable to self-report: A systematic review of psychometric properties. *Dev Med Child Neurol*. 2023; **65**(8): 1029-42.
211. Smith MG, Gibson RJ, Russo RN, Karanicolas S, Harvey AR. Examining tools for assessing the impact of chronic pain on emotional functioning in children and young people with cerebral palsy: stakeholder preference and recommendations for modification. *Quality of Life Research*. 2024; **33**(8): 2247-59.
212. Stansbury J, Crary M, Bangert L, Root J, Bolf K, Jefferis K, Barney C. Standardized clinical assessment of chronic pain in children and adults with intrathecal baclofen pumps. *Developmental Medicine And Child Neurology*. 2017; **59**(Supplement 3): 99.
213. Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007; **19**(6): 349-57.
214. Palinkas LA, Horwitz SM, Green CA, Wisdom JP, Duan N, Hoagwood K. Purposeful Sampling for Qualitative Data Collection and Analysis in Mixed Method Implementation Research. *Administration and policy in mental health and mental health services research*. 2013; **42**(5): 533-44.
215. Harris PA, Taylor R, Thielke R, Payne J, Gonzalez N, Conde JG. Research electronic data capture (REDCap)—A metadata-driven methodology and workflow process for providing translational research informatics support. *J Biomed Inform*. 2009; **42**(2): 377-81.
216. Harris PA, Taylor R, Minor BL, Elliott V, Fernandez M, O'Neal L, et al. The REDCap consortium: Building an international community of software platform partners. *J Biomed Inform*. 2019; **95**: 103208.
217. Smart A. A multi-dimensional model of clinical utility. *Int J Qual Health Care*. 2006; **18**(5): 377-82.
218. Zempsky WT, O'Hara EA, Santanelli JP, Palermo TM, New T, Smith-Whitley K, Casella JF. Validation of the sickle cell disease pain burden interview-youth. *J Pain*. 2013; **14**(9): 975-82.
219. Simons LE, Smith A, Ibagon C, Coakley R, Logan DE, Schechter N, et al. Pediatric Pain Screening Tool: rapid identification of risk in youth with pain complaints. *PAIN*. 2015; **156**(8): 1511-8.
220. Eccleston C, Jordan A, McCracken LM, Sleed M, Connell H, Clinch J. The Bath Adolescent Pain Questionnaire (BAPQ): development and preliminary psychometric evaluation of an instrument to assess the impact of chronic pain on adolescents. *PAIN*. 2005; **118**(1-2): 263-70.
221. McCracken LM. Behavioral constituents of chronic pain acceptance: Results from factor analysis of the chronic pain acceptance questionnaire. *J Back Musculoskelet Rehabil*. 1999; **13**(2-3): 93-100.
222. Heathcote LC, Bhandari RP, Timmers I, Harrison LE, Simons LE. Rapid identification and clinical indices of fear-avoidance in youth with chronic pain. *Pain (Amsterdam)*. 2020; **161**(3): 565-73.
223. McCracken LM. “Attention” to pain in persons with chronic pain: A behavioral approach. *Behav Ther*. 1997; **28**(2): 271-84.

224. Crombez G, Bijttebier P, Eccleston C, Mascagni T, Mertens G, Goubert L, Verstraeten K. The child version of the pain catastrophizing scale (PCS-C): a preliminary validation. *Pain (Amsterdam)*. 2003; **104**(3): 639-46.
225. Bradshaw C, Atkinson S, Doody O. Employing a Qualitative Description Approach in Health Care Research. *Global qualitative nursing research*. 2017; **4**: 2333393617742282-.
226. Jack SM, Phoenix M. Qualitative health research in the fields of developmental medicine and child neurology. *Dev Med Child Neurol*. 2022; **64**: 830–9.
227. Kallio H, Pietilä A-M, Johnson M, Kangasniemi M. Systematic methodological review: developing a framework for a qualitative semi-structured interview guide. *Journal of Advanced Nursing*. 2016; **72**(12): 2954-65.
228. Vears D, Gillam L. Inductive content analysis: A guide for beginning qualitative researchers. *Focus on Health Professional Education*. 2022; **23**(1).
229. Murphy J, Cameron L. The effectiveness of Talking Mats with people with intellectual disability. *Br J Learn Disabil*. 2008; **36**(4): 232-41.
230. Headrick K, Thornton M, Hogan A, Deramore Denver B, Drake G, Wallen M. Consumer involvement in research – parent perceptions of partnership in cerebral palsy research: a qualitative study. *Disability And Rehabilitation*. 2023; **45**(3): 483-93.
231. Niederberger M, Köberich S. Coming to consensus: the Delphi technique. *Eur J Cardiovasc Nurs*. 2021; **20**(7): 692-5.
232. Spranger J, Homberg A, Sonnberger M, Niederberger M. Reporting guidelines for Delphi techniques in health sciences: A methodological review. *Z Evid Fortbild Qual Gesundhwes*. 2022; **172**: 1-11.
233. Harvey AR, McKinnon CT, Smith N, Ostojic K, Paget SP, Smith S, et al. Establishing consensus for the assessment of chronic pain in children and young people with cerebral palsy: a Delphi study. *Disability And Rehabilitation*. 2021; **44**(23): 7161–6.
234. McGrath PJ, Walco GA, Turk DC, Dworkin RH, Brown MT, Davidson K, et al. Core Outcome Domains and Measures for Pediatric Acute and Chronic/Recurrent Pain Clinical Trials: PedIMMPACT Recommendations. *The Journal of Pain*. 2008; **9**(9): 771-83.
235. Heathcote LC, Bhandari RP, Timmers I, Harrison LE, Simons LE. Rapid identification and clinical indices of fear-avoidance in youth with chronic pain. *PAIN*. 2020; **161**(3): 565-73.
236. Simons LE, Sieberg CB, Carpino E, Logan D, Berde C. The Fear of Pain Questionnaire (FOPQ): Assessment of Pain-Related Fear Among Children and Adolescents With Chronic Pain. *J Pain*. 2011; **12**(6): 677-86.
237. Cleeland CS, Ryan KM. Pain assessment: global use of the Brief Pain Inventory. *Ann Acad Med Singap*. 1994; **23**(2): 129-38.
238. Engel JM, Kartin D, Carter GT, Jensen MP, Jaffe KM. Pain in youths with neuromuscular disease. *Am J Hosp Palliat Care*. 2009; **26**(5): 405-12.
239. Keeney S, Hasson F, McKenna HP. A critical review of the Delphi technique as a research methodology for nursing. *Int J Nurs Stud*. 2001; **38**(2): 195-200.
240. Terwee CB, Prinsen CAC, Chiarotto A, Westerman MJ, Patrick DL, Alonso J, et al. COSMIN methodology for evaluating the content validity of patient-reported outcome measures: a Delphi study. *Quality of Life Research*. 2018; **27**(5): 1159-70.
241. Murphy MK, Black NA, Lamping DL, McKee CM, Sanderson CF, Askham J, Marteau T. Consensus development methods, and their use in clinical guideline development. *Health Technol Assess*. 1998; **2**(3): i-iv.

242. Keeney S, Hasson F, McKenna HP. The Delphi technique in nursing and health research. Chichester, West Sussex: Wiley-Blackwell; 2011.
243. Diamond IR, Grant RC, Feldman BM, Pencharz PB, Ling SC, Moore AM, Wales PW. Defining consensus: A systematic review recommends methodologic criteria for reporting of Delphi studies. *J Clin Epidemiol*. 2014; **67**(4): 401-9.
244. Mokkink LB, Elsman EBM, Terwee CB. COSMIN guideline for systematic reviews of patient-reported outcome measures version 2.0. *Qual Life Res*. 2024.
245. Emmott A, West N, Zhou G, Dunsmuir D, Montgomery CJ, Lauder GR, von Baeyer CL. Validity of Simplified Versus Standard Self-Report Measures of Pain Intensity in Preschool-Aged Children Undergoing Venipuncture. *J Pain*. 2017; **18**(5): 564-73.
246. Schmidt AK, van Gorp M, van Wely L, Ketelaar M, Hilberink SR, Roebroeck ME, Groups P-DPS. Autonomy in participation in cerebral palsy from childhood to adulthood. *Dev Med Child Neurol*. 2020; **62**(3): 363-71.
247. Matza LS, Patrick DL, Riley AW, Alexander JJ, Rajmil L, Pleil AM, Bullinger M. Pediatric Patient-Reported Outcome Instruments for Research to Support Medical Product Labeling: Report of the ISPOR PRO Good Research Practices for the Assessment of Children and Adolescents Task Force. *Value Health*. 2013; **16**(4): 461-79.
248. Kramer JM, Schwartz A. Reducing Barriers to Patient-Reported Outcome Measures for People With Cognitive Impairments. *Arch Phys Med Rehabil*. 2017; **98**(8): 1705-15.
249. Hasson F, Keeney S, McKenna H. Research guidelines for the Delphi survey technique. *J Adv Nurs*. 2000; **32**(4): 1008-15.
250. von Baeyer CL, Jaaniste T, Vo HLT, Brunsdon G, Lao H-C, Champion GD. Systematic Review of Self-Report Measures of Pain Intensity in 3- and 4-Year-Old Children: Bridging a Period of Rapid Cognitive Development. *The Journal of Pain*. 2017; **18**(9): 1017-26.
251. Kasparova P, Greaves J. Validity of simplified self-report methods versus standard use method for pain assessment in preschool-aged children. *Journal for Specialists in Pediatric Nursing*. 2023; **28**(3): e12411-n/a.
252. Vedel I, Kaur N, Hong QN, El Sherif R, Khanassov V, Godard-Sebillotte C, et al. Why and how to use mixed methods in primary health care research. *Fam Pract*. 2019; **36**(3): 365-8.
253. Kramer JM, Schwartz A. Refining the Pediatric Evaluation of Disability Inventory–Patient-Reported Outcome (PEDI-PRO) item candidates: interpretation of a self-reported outcome measure of functional performance by young people with neurodevelopmental disabilities. *Developmental Medicine & Child Neurology*. 2017; **59**(10): 1083-8.
254. Murphy J, Gray CM, van Achterberg T, Wyke S, Cox S. The effectiveness of the Talking Mats framework in helping people with dementia to express their views on well-being. *Dementia (London, England)*. 2010; **9**(4): 454-72.
255. Patrick DL, Burke LB, Gwaltney CJ, Leidy NK, Martin ML, Molsen E, Ring L. Content Validity—Establishing and Reporting the Evidence in Newly Developed Patient-Reported Outcomes (PRO) Instruments for Medical Product Evaluation: ISPOR PRO Good Research Practices Task Force Report: Part 2—Assessing Respondent Understanding. *Value in Health*. 2011; **14**(8): 978-88.
256. Hak T, Veer Kvd, Jansen H. The Three-Step Test-Interview (TSTI): An observation-based method for pretesting self-completion questionnaires. *Survey Research Methods*. 2008; **2**(3): 143-50.

257. Teachman G. Optimizing Interviews with Children and Youth with Disability. In: Liamputtong P, editor. Handbook of Research Methods in Health Social Sciences. Singapore: Springer Singapore; 2019. p. 2023-40.
258. Brédart A, Marrel A, Abetz-Webb L, Lasch K, Acquadro C. Interviewing to develop Patient-Reported Outcome (PRO) measures for clinical research: Eliciting patients' experience. *Health and Quality of Life Outcomes*. 2014; **12**(1): 15.
259. Walsh M, Harman I, Manning P, Ponza B, Wong S, Shaw B, et al. Including People Who Use Augmentative and Alternative Communication in Qualitative Research: Can You Hear Us? *International journal of qualitative methods*. 2024; **23**.
260. Hsieh H-F, Shannon SE. Three Approaches to Qualitative Content Analysis. *Qualitative health research*. 2005; **15**(9): 1277-88.
261. Rinde ER, Anderzén-Carlsson A, Jahnsen RB, Andersen RD. "Pain is one piece of a complex jigsaw puzzle" – experiences of raising a child with cerebral palsy who has pain. *Disability And Rehabilitation*. 2023; **46**(20): 4763–71.
262. Lindsay S. Perceptions of health care workers prescribing augmentative and alternative communication devices to children. *Disability and rehabilitation: Assistive technology*. 2010; **5**(3): 209-22.
263. Yau SH, Choo K, Tan J, Monson O, Bovell S. Comparing and contrasting barriers in augmentative alternative communication use in nonspeaking autism and complex communication needs: multi-stakeholder perspectives. *Front Psychiatry*. 2024; **15**: 1385947.
264. Eriksson E, Hägglund G, Alriksson-Schmidt AI. Pain in children and adolescents with cerebral palsy - a cross-sectional register study of 3545 individuals. *BMC Neurology*. 2020; **20**(1): 1-9.
265. Hunt A, Goldman A, Seers K, Crichton N, Mastroyannopoulou K, Moffat V, et al. Clinical validation of the paediatric pain profile. *Dev Med Child Neurol*. 2004; **46**(1): 9-18.
266. Raskoff SZ, Thurm A, Miguel HO, Kim SYH, Quezado ZMN. Pain research and children and adolescents with severe intellectual disability: ethical challenges and imperatives. *The Lancet Child & Adolescent Health*. 2022; **7**(4): 288-96.
267. Mimmo L, Woolfenden S, Travaglia J, Strnadová I, Phillips MT, Karen, et al. Codesigning patient experience measures for and with children and young people with intellectual disability: a study protocol. *BMJ Open*. 2021; **11**(12): e050973.
268. Smith M, Russo R, Gibson R, Schibani M, Harvey A. D42. Development and testing of adapted versions of two chronic pain assessment tools for young people with cerebral palsy (CP): AusACPDM 2024 Abstracts:. *Developmental Medicine & Child Neurology*. 2024; **66**(S3): 23.
269. Smith MG, Gibson RJ, Russo RN, Harvey AR. Adapting two pain assessment tools for young people with cerebral palsy: a multi-stakeholder consensus study. *Pain Rep*. 2025; **10**(4): e1304.
270. Smith MG, Gibson RJ, Schibani M, Russo RN, Thirumanickam A, Harvey AR. The comprehensibility and feasibility of the modified brief pain inventory and fear of pain questionnaire adapted for children and young people with cerebral palsy. *Quality of Life Research*. 2025.
271. Mokkink LB, Terwee CB, Patrick DL, Alonso J, Stratford PW, Knol DL, et al. The COSMIN study reached international consensus on taxonomy, terminology, and definitions of measurement properties for health-related patient-reported outcomes. *J Clin Epidemiol*. 2010; **63**(7): 737-45.

272. Crombez G, Eccleston C, Van Damme S, Vlaeyen JWS, Karoly P. Fear-Avoidance Model of Chronic Pain: The Next Generation. *Clin J Pain*. 2012; **28**(6): 475-83.
273. Vlaeyen JWS, Crombez G, Linton SJ. The fear-avoidance model of pain. *PAIN*. 2016; **157**(8): 1588-9.
274. R Core Team. R: A Language and Environment for Statistical Computing. Vienna, Austria2021.
275. de Vet HCW, Adèr HJ, Terwee CB, Pouwer F. Are Factor Analytical Techniques Used Appropriately in the Validation of Health Status Questionnaires? A Systematic Review on the Quality of Factor Analysis of the SF-36 [with Commentary and Response]. *Qual Life Res*. 2005; **14**(5): 1203-24.
276. Rosseel Y. lavaan: An R Package for Structural Equation Modeling. *J Stat Softw*. 2012; **48**(2): 1 - 36.
277. Hu L-t, Bentler PM. Cutoff criteria for fit indexes in covariance structure analysis: Conventional criteria versus new alternatives. *Struct Equ Modeling*. 1999; **6**(1): 1-55.
278. Crutzen R, Peters G-JY. Scale quality: alpha is an inadequate estimate and factor-analytic evidence is needed first of all. *Health Psychol Rev*. 2017; **11**(3): 242-7.
279. Coster WJ, Ni P, Slavin MD, Kisala PA, Nandakumar R, Mulcahey MJ, et al. Differential item functioning in the Patient Reported Outcomes Measurement Information System Pediatric Short Forms in a sample of children and adolescents with cerebral palsy. *Dev Med Child Neurol*. 2016; **58**(11): 1132-8.
280. Hristova M, Manolova H, Staykova S, Markova G. Assessment of Intellectual Functioning among Children with Neurodevelopmental Disorders: Challenges and Implications Beyond the Clinical Practice. *J Intellect Dev Disabil*. 2018; **5**(4): 124-9.
281. Smith NL, Gibson N, Imms C, Thornton AL, Harvey AR. A core outcome set to assess chronic pain interference and impact on emotional functioning for children and young people with cerebral palsy. *Developmental Medicine & Child Neurology*. 2025; **n/a**(n/a).
282. Chambers CT. From evidence to influence: dissemination and implementation of scientific knowledge for improved pain research and management. *PAIN*. 2018; **159 Suppl 1**: S56-s64.
283. Walsh M, Harman I, Manning P, Ponza B, Wong S, Shaw B, et al. Including People Who Use Augmentative and Alternative Communication in Qualitative Research: Can You Hear Us? *International journal of qualitative methods*. 2024; **23**: 16094069241234190.
284. Cook KF, Bamer AM, Roddey TS, Kraft GH, Kim J, Amtmann D. A PROMIS fatigue short form for use by individuals who have multiple sclerosis. *Quality of Life Research*. 2012; **21**(6): 1021-30.
285. Meirte J, Tyack Z. Electronic Patient-Reported Outcome Measures in Burn Scar Rehabilitation: A Guide to Implementation and Evaluation. *European Burn Journal*. 2022; **3**(2): 290-308.
286. Graham ID, Logan J, Harrison MB, Straus SE, Tetroe J, Caswell W, Robinson N. Lost in knowledge translation: Time for a map? *Journal of Continuing Education in the Health Professions*. 2006; **26**(1): 13-24.
287. Stewart K, de Vries T, Harvey A. Implementing accurate identification and measurement of dyskinesia in cerebral palsy into clinical practice: A knowledge translation study. *Journal of Paediatrics and Child Health*. 2019; **55**(11): 1351-6.
288. Gannotti ME, Gross PH, Thorpe DE, Hurvitz EA, Noritz GH, Horn SD, et al. Cerebral palsy research network community registry adult surveys on function & pain:

Successes, challenges, and future directions. *Disability and Health Journal*. 2024: 101625.
289. Anthony SJ, Pol SJ, Lin J, Barwick M, Brudno M, Manase D, et al. Creation of an electronic patient-reported outcome measure platform Voxe: a mixed methods study protocol in paediatric solid organ transplantation. *BMJ Open*. 2021; **11**(10): e053119.
290. Anthony SJ, Pol SJ, Selkirk EK, Matthiesen A, Klaassen RJ, Manase D, et al. User-Centered Design and Usability of Voxe as a Pediatric Electronic Patient-Reported Outcome Measure Platform: Mixed Methods Evaluation Study. *JMIR Hum Factors*. 2024; **11**: e57984.
291. Higgins KS, Tutelman PR, Chambers CT, Witteman HO, Barwick M, Corkum P, et al. Availability of researcher-led eHealth tools for pain assessment and management: barriers, facilitators, costs, and design. *Pain Rep*. 2018; **3**(Suppl 1): e686.
292. Vinkel MN, Rackauskaite G, Østergaard JR, Finnerup NB, Jensen MP. Pain coping and catastrophizing in youth with and without cerebral palsy. *Scandinavian Journal of Pain*. 2024; **24**(1).
293. Liossi C, Johnstone L, Lilley S, Caes L, Williams G, Schoth DE. Effectiveness of interdisciplinary interventions in paediatric chronic pain management: a systematic review and subset meta-analysis. *British journal of anaesthesia : BJA*. 2019; **123**(2): e359-e71.
294. Smith N, Garbellini S, Bear N, Thornton A, Watts P, Gibson N. Effect of targeted movement interventions on pain and quality of life in children with dyskinetic cerebral palsy: a pilot single subject research design to test feasibility of parent-reported assessments. *Disabil Rehabil*. 2023; **45**(10): 1646-54.
295. Stinson JN, Lalloo C, Harris L, Isaac L, Campbell F, Brown S, et al. iCanCope with Pain™: User-centred design of a web- and mobile-based self-management program for youth with chronic pain based on identified health care needs. *Pain Res Manag*. 2014; **19**(5): 257-65.
296. Slater H, Stinson JN, Jordan JE, Chua J, Low B, Lalloo C, et al. Evaluation of Digital Technologies Tailored to Support Young People's Self-Management of Musculoskeletal Pain: Mixed Methods Study. *J Med Internet Res*. 2020; **22**(6): e18315.
297. White R. Media: painHEALTH: an evidence-based website for adult consumers experiencing musculoskeletal pain. *J Physiother*. 2018; **64**(3): 202-3.
298. Rinde ER, Anderzén-Carlsson A, Jahnsen RB, Andersen RD. Children with Cerebral Palsy and Their Parents Have Different Experiences of Pain Management: A Qualitative Study. *Children*. 2024; **11**(9): 1055.
299. King G, Chiarello L. Family-Centered Care for Children With Cerebral Palsy: Conceptual and Practical Considerations to Advance Care and Practice. *Journal of Child Neurology*. 2014; **29**(8): 1046-54.
300. Beveridge JK, Noel M, Soltani S, Neville A, Orr SL, Madigan S, Birnie KA. The association between parent mental health and pediatric chronic pain: a systematic review and meta-analysis. *Pain (Amsterdam)*. 2024; **165**(5): 997.
301. Brandelli YN, Tutelman PR, Chambers CT, Parker JA, Stinson JN, Huber AM, et al. "Every Little Furrow of Her Brow Makes Me Want To Stop": An Interpretative Phenomenologic Analysis of Mothers' Experiences With Juvenile Idiopathic Arthritis Treatments. *Arthritis Care Res (Hoboken)*. 2022; **74**(11): 1761-9.
302. Wallwork SB, Shenk C, McMurtry CM, Hood AM, Pavlova M, Olson AE, et al. “I hear you”. Validation in the context of children's pain as an untapped opportunity to prevent chronic pain. *Pain (Amsterdam)*. 2024.

Talking Mats® copyright statement

Talking Mats® is a registered trademark. The term ‘Talking Mats’ cannot be used commercially by other organisations.

The Symbols are designed and © to Adam Murphy 2015 and assigned to Talking Mats Ltd. In perpetuity. They may not be reproduced without permission.

Talking Mats® was developed at the University of Stirling and the intellectual property rights in Talking Mats are owned by the University of Stirling and utilised by Talking Mats Limited under exclusive licence.

Training in Talking Mats must be delivered by an accredited trainer who holds a current licence with Talking Mats Ltd.