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Review Article

The supportive care needs of parents caring for a child with a rare disease: A scoping review

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Abstract

Background: Parents caring for a child with a rare disease report unmet needs, the origins of which are varied and complex. Few studies have systematically attempted to identify the supportive care needs of parents with a child with a rare disease comprehensively. We have used the widely accepted Supportive Care Needs Framework (SCNF) as the structure for this review.

Objective: The purpose of the current review was to identify the supportive care needs of parents with a child with a rare disease, irrespective of condition.

Methods: We conducted a scoping study review comprising 29 studies (1990–2014) to identify and examine the research literature related to the supportive care needs of parents, and to compare these needs with the seven domains outlined in the SCNF.

Results: Most common needs cited were social needs (72% of papers), followed by informational needs (65% of papers) and emotional needs (62% of papers), with the most common parental needs overall being information about their child's disease, emotional stress, guilt and uncertainty about their child's future health care needs, parents own caring responsibilities and the need for more general support.

Conclusion: A paucity of studies exists that explore the supportive care needs of parents of a child with a rare disease. The SCNF only partially reflects the breadth and type of needs of these parents, and a preliminary revised framework has been suggested. Further research is required in this area, particularly empirical research to amend or confirm the suggested new framework.

Keywords: Parents; Supportive care needs; Rare diseases; Impact; Framework

Rare diseases, including those of non-genetic origin, are defined as complex, multi-systemic, often life-threatening, incurable and non-preventable diseases.[1] Rare diseases, by nature have a low prevalence. However, low prevalence does not mean low impact, for when combined, rare diseases affect a significant percentage of the total population.[2,3] Collectively, rare diseases affect between 6 and 10% of the global population.[4,5] In Europe, a rare disease is defined as one affecting less than 1/2000 individuals, in the USA it is fewer than 200,000, whilst in Australia it is 1/10000.[3] There exist between 6000 and 8000 distinct rare diseases, many of which have no formal title and are difficult to diagnose.[6,7] Around 75% of rare diseases affect a child's quality of life from birth.[8] Because of the rarity, the medical, scientific and political communities have typically neglected parents because they are considered a diffuse minority.[6,9] For many of the parents of a child with a rare disease, the burden of care spans many years and involves a lifetime commitment. It often requires a change in work patterns, income and domestic responsibilities. Parents will require specialist health literacy, care giving skills and resources beyond those normally required by parents in general. Although parents of a child with a chronic health condition face many similar issues, parenting a child with a rare disease has added difficulties because diagnosis may be delayed or undetermined, support groups may be small and geographically scattered and health care skills and resources limited.[5,10] Importantly, the symptoms and needs of the affected children are likely to be heterogenous, whereas their parents' supportive care needs are likely to be homogenous. Identification of the supportive care needs of parents with a child with a rare disease has not previously been

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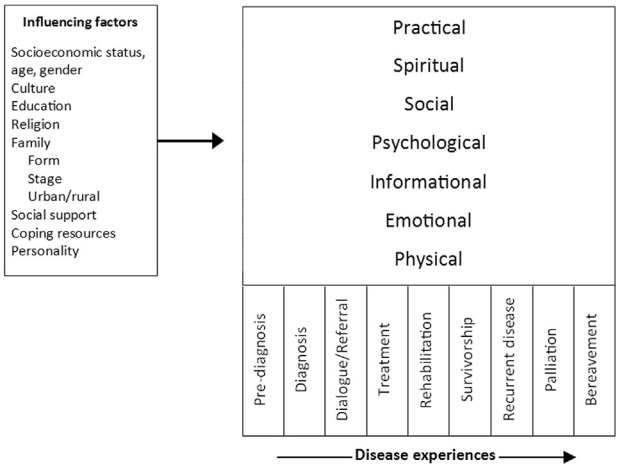
systematically attempted and therefore initiatives to provide appropriate support have presumably until now been ad hoc.

A major issue for parents is that early detection and definitive diagnosis is often difficult.[10,11] In contrast, more common diseases are identified during early stages of pregnancy and at birth (e.g. Down syndrome and Cystic fibrosis), making support care pathways more determined.

The literature discussing the needs of parents caring for a child with a rare disease most often only relate to a specific disease under study, are largely based on small sample sizes, and are often limited to a particular country/culture making it difficult to generalize to parents of children with other diseases. Up to the present time, few studies exist that have attempted to identify the supportive care needs of parents in a comprehensive fashion, either as a review or as an original study.[12,13] One of the exceptions is an Australian study by Anderson et al[4] that did look at parents of children with any rare inherited metabolic disease. However, this study was based on existing questionnaires, with no attempt to explore needs not already identified in these instruments. To date, almost all research into supportive care has focussed on the affected individual, with little recognition of the burden that this has imposed on the parents. In the case of rare diseases, this is a particularly important omission. There currently does not exist a conceptual framework that outlines a taxonomy of supportive care needs of parents with a child with a rare disease, irrespective of the condition.



Conceptual view of the supportive care needs and influencing factors of individuals living with cancer along their spectrum of disease (Fitch, 1994)

Fig. 1. The supportive care needs framework.

Supportive care

Supportive care is described as "the provision of necessary services as defined by those living with or affected by cancer to meet their physical, psychosocial, informational and spiritual needs during the pre-diagnostic, diagnostic, treatment, and follow-up phases, while encompassing issues of survivorship, palliation, and bereavement" [Fitch 1994 in 13]. In 2000, a Supportive Care Needs Framework (SCNF) was developed at the Cancer Care Ontario foundation in Canada to help guide health professionals to ensure the supportive care needs of cancer patients are being met within all domains of care, and along the disease continuum.[14] Each domain of care is thought to vary across a continuum, differing in level of priority depending on the unique needs of the cancer patient.[12] The SCNF consists of seven domains of need, each influenced by external factors such as, SES, age, gender, culture, education, family, social support and personality (see Fig. 1). The seven domains of supportive care needs include; practical, spiritual, social, psychological, informational, emotional and physical needs. Since its development, the SCNF has been widely accepted and referred to across numerous studies investigating unmet supportive care needs of oncology groups and their family members living with cancer.[14–17]

In later studies by Kerr and colleagues[12,13] who sought to evaluate the SCNF as a means of describing and categorizing specific needs of parents of children with cancer, admitted that the framework had issues in classifying the supportive care needs of these parents, including many needs that fell into more than one domain. Despite its recognized flaws, the SCNF is one of the most widely used frameworks for supportive care needs, and thus we have used it for the structure of this review.

Study objective

The objectives of this scoping review are: (i) to gather and appraise all available published literature which investigates parental supportive care needs of a child with a rare disease; (ii) to compare and contrast these needs with the seven domains outlined in the SCNF; and (iii) to determine whether a new supportive care needs framework is required for parents with a child with a rare disease.

Review methodology

The methodologically rigorous scoping study, as proposed by Arksey and O'Malley,[18] was chosen for this review to conduct a comprehensive and systematic search of the literature to establish the full range and nature of the supportive care needs of parents caring for a child with a rare disease. This review follows the six-stage scoping review framework outlined by Arksey and O'Malley.[18] The sixth stage, considered optional, was not included in this review.

Stage one: identify the research question

A two-part research question was developed: (i) What are the supportive care needs of parents caring for a child diagnosed with a rare disease; and (ii) Can these supportive

care needs be satisfactorily grouped under the existing SCNF classifications?

Stage two: identify relevant studies

For a comprehensive search of the literature, relevant peer-reviewed journal articles published between 1990 and April 2014 were sought from the following eight electronic databases; MEDLINE, EMBASE, CINAHL, Psych INFO, Health Sources: Nursing/Academic Edition, Cochrane database, Scopus, Web of Science. Key search terms and word combinations included; (parent* or father* or mother*) and (support* or need* or issue* or problem* or impact*) and (rare) and (supportive care need* or supportive care) and (rare disorder* or rare disease* or rare condition* or neglected disease* or orphan disease* or chronic illness* or chronic condition*) and (disorder* adj3 rare or disease* adj3 rare or condition* adj3 rare). In addition, a hand search of the reference lists of selected articles was undertaken and the use of Google Scholar to access any other primary sources and full text versions of articles was utilized. We exported all literature sources from each of the database searches into a references and bibliographic management software program.

Stage three: study selection

The inclusion criteria were; (1) articles written in English; (2) published original peer-reviewed journals; (3) those with an approved ethics statement; (4) a focus exploring or identifying the support needs of parents with a child with a rare disease; and (5) the population sample included parents; mothers and/or fathers. For the purposes of this review, we have defined a disease as rare if it has a low prevalence, occurs infrequently and affects a limited number of people in the general population at any one time.[19] Further, although we have used the phrase ''rare disease'', we have taken a broad concept of disease to refer to any impairment of health, whether physical or mental. Articles that were considered poorly conducted, too obscure/psychologically focused, extremely small sample sizes and minimal or no analysis of findings; and studies that were considered editorials, discussion points or personal opinion pieces were excluded. A second author (AE) reviewed each paper with the lead author (LP) to decide whether it should be included. Using the above key search descriptors, 156 articles were identified across the eight databases. Of those, 29 were included in this review (see Fig. 2).

Stage four: charting the data

The fourth stage involved developing a framework for charting data. A summarizing process, in a standardized manner, was executed as described by Arksey and O'Malley.[18] A data charting form was developed by the researcher summarizing each primary reviewed article by; author, year, country of origin, study aim, childhood disease, study design, study methods and sample size, an abridged summary of the findings and limitations for each study, and parental needs identified based on the SCNF framework were listed. A second reviewer (AE) then validated the data by reviewing each selected article based on the inclusion/exclusion criteria; a consensus was made of the information presented in Table 1.

Stage five: collating, summarizing and reporting results

This fifth and final stage of the review involved collating descriptions of supportive care needs into themes. We based the development of the themes on a low inference simple qualitative descriptive approach recommended by Sandelowski[20] and then used these themes to summarize supportive care needs emerging from the contemporary literature under review. Reporting consisted of a comparison of the scoping reviewing findings with the adequacy of concepts outlined in the SCNF.

Results

A total of 29 studies met the inclusion criteria (see Table 1). Four studies used quantitative methods, 18 used qualitative, three used a mixed methods approach, and four were literature reviews. In each of the quantitative studies, questionnaires ($n = 8$) were used to gather data; in the qualitative studies, face-to-face ($n = 15$), focus groups ($n = 1$) and telephone ($n = 2$) interviews were conducted to gather data. The mixed methods studies used a combination of interviews and questionnaires. Four literature review papers were examined spanning 1992–2009, and referencing 102 papers.

Of the 29 studies reviewed, parental needs were cited across all seven domains of the SCNF, with the majority ($n = 18$) citing three or more SCN domains. Most common was social needs ($n = 21$; 72%), followed by informational needs ($n = 19$; 65%) and emotional needs ($n = 18$; 62%). In the sections that follow, the needs are discussed in the order of how often they were cited in the literature. However, we note that this does not necessarily imply importance. Further, domains of supportive care needs do not appear to be mutually exclusive with many needs that could fit into more than one domain.

Social needs

Of the social needs cited, parents most frequently voiced the need for more support in all areas.[4,11,12,21–25] Access to a support group was considered by parents as an important means of support for them.[4,12,23,26,27] Family and friends were also an important source of support.[12,22,24] Parents felt that as time passed, support offered by others became less, even though their circumstances had not changed.[24]

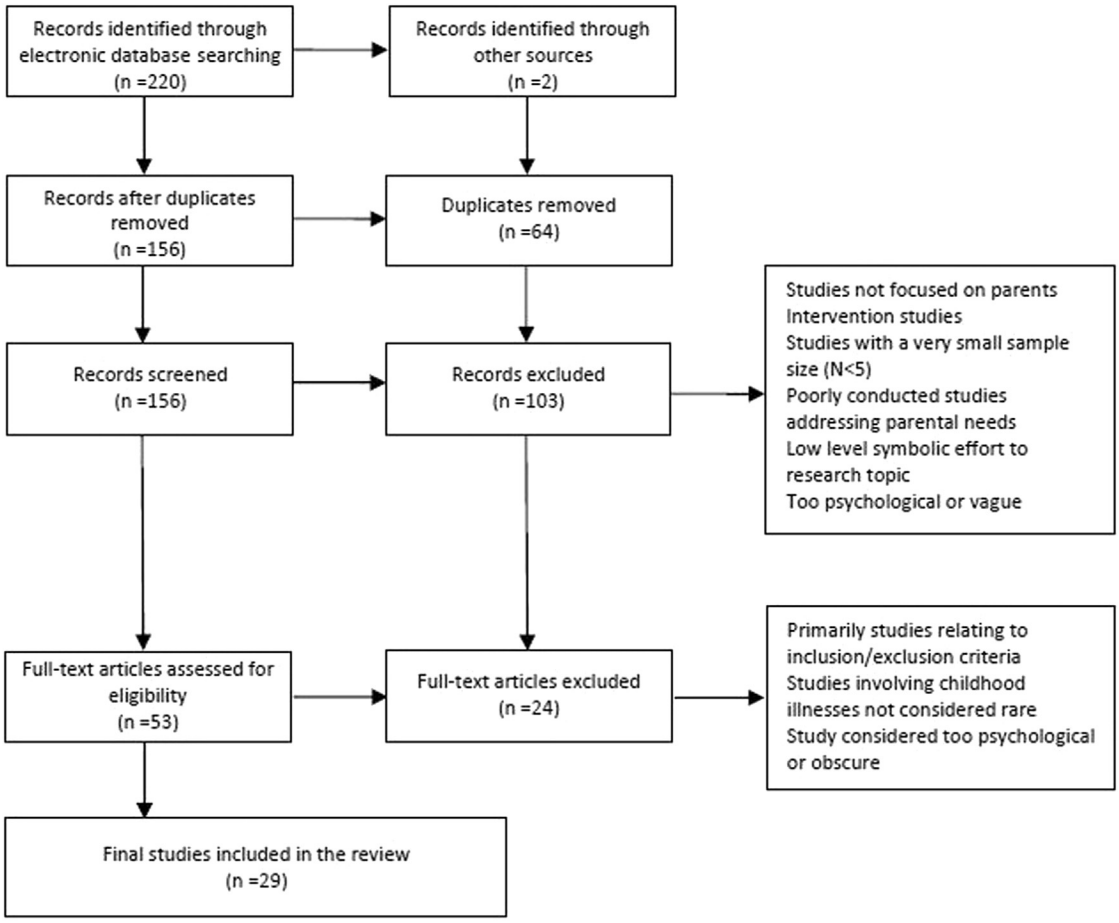


Fig. 2. PRISMA flow diagram of scoping literature search and selection.

Some parents went to great lengths to access necessary supports, including looking overseas.[4]

Communicating with other parents was the second most frequently cited social need.[12,21,23,24,27–29] Parents felt that only other parents in similar circumstances to them could truly understand, and that by talking to these parents it would enable them to better cope and have a more positive outlook on life.[28,29] It was also important for parents to communicate with others who were not involved in the direct medical care of their child.[21]

Social isolation, loneliness and feeling disconnected because of their child's disease and their caring routines were common issues cited by parents.[21,22,25,28–30] Parents felt that their social life was lacking, there was a loss of freedom, and they longed for more spontaneity.[22,28,30–33] Some parents described their social situation as "having no life".[28] For some parents, the complexity of their child's disease, relying on cumbersome medical equipment, meant their ability to attend social activities was limited.[31] Avoiding certain social situations in order to avoid having to deal with public embarrassment and humiliation only further compounded feelings of social isolation in parents.[29,32,34]

Almost all relationships were adversely affected as a result of caring responsibilities.[11,23,30–32,35] The daily burden of care and missed opportunities to spend quality time with each other had a negative impact on partner relationships.[11,30–32] Support for and fear of neglecting the other siblings of the sick child was also a significant concern for parents.[11,32,35]

Finally, parents found balancing work and family challenging.[32,33,36] Some parents, particularly mothers, had to take time off work or altogether leave their place of employment to care for their child, and career ambition and personal interests had to be set aside in order to focus on their child's caring needs.[32,33,36]

Informational needs

Parents most frequently reported the need for general medical information regarding their child's disease.[4,12,13,21,23,37–40] There was a strong projection of needs; parents wanted information concerning their child's future health and life chances. They wanted to know what impacts their child's disease and prescribed medications would have on the child's development, learning and long-term health.[21,23,39] They also desired information on what long-term services and community health supports were available for their child as they developed, so that they could more confidently plan for the future.[11,23,26,39]

Table 1
Details of primary studies identified and reviewed

Author (year)	Country	Study aim	Disease	Study design/methods/sample	Findings/critique of study	Domains of SCNF
Anderson, Elliott, and Zurynski (2013)	AUS	To assess the health, psychosocial and financial impacts of Australian families	Rare disorders	Quantitative study/online questionnaire based on five established surveys/n = 30 parents (24 mothers, 6 fathers)	Need more information regarding child's disorder. Few parents received psychological counselling/support despite feeling distressed. Around time of diagnosis is an important period for assessing parental psychological needs. Financial problems reported. Difficulties accessing social supports. Small sample size. New questions tested for validity but not reliability. Fathers under sampled	Informational, practical, psychological and social needs
Aytch, Hammond and White (2001)	USA	To identify perceived needs of parents for information, resources and support in caring for their child	Seizures	Mixed methods study/face-to-face open-ended interviews/lasting 1.5−2 h/questionnaire administered at time of interviews/participants were recruited from a University medical center pediatric neurology clinic/n = 31 mothers, 29 fathers	Need more information regarding effects of seizures and medications on child's development/long-term health. Need for education/resources to provide family and friends. Parents felt overwhelmed by volume of information available; majority considered not relevant to them. Emotional needs not addressed by health professionals. Parents felt disconnected from health and community services. Desired to communicate with other parents. A well conducted study. Questionnaire not tested for validity or reliability	Informational, emotional and social needs
Brewer et al (2008)	UK	To describe the experiences of parents caring for their child or younger person	Juvenile Huntington's disease	Qualitative study/semi-structured interviews/lasting 45minutes-3hours/participants were recruited by National JHD Register/n = 12 parents (8 mothers, 4 fathers)	Health professionals lack knowledge of condition. Parents having to assume the role of 'expert'. Lack of information available to parents hindered their ability to manage situations. Parents felt isolated. Difficulty in asking for help/support. Parents would avoid certain social situations that were potentially unpleasant/embarrassing which further increased their sense of isolation. Aim of study not made clear	Informational and social needs
Coffey (2006)	USA	To provide a metasynthesis of qualitative literature pertaining to parenting a child with a chronic illness	Chronic illness	Review of the literature/articles published in English 1989−2000/n = 11 articles included in review	Daily emotional needs and feelings of parents caring for their child discussed. Parents concerned about their child's future health and wellbeing. Also concerned for the impact that chronic illness might have on the siblings. Parents describe feeling isolated, having a loss of freedom and a lack of support systems.	Emotional and social needs

(*Continued*)

Table 1
Continued

Author (year)	Country	Study aim	Disease	Study design/methods/sample	Findings/critique of study	Domains of SCNF
					The search strategy was well defined. Only qualitative studies were considered	
Duffy (2011)	USA	To critically analyze current literature related to psychological and emotional challenges of parents	Epilepsy	Review of the literature/articles published in English 1995−2009/n = 34 articles included in review	Parental stress, worry and fear related to child with epilepsy. Level of anxiety and stress influenced by severity of disease amongst parents. Need for social and family support. Need for information/knowledge of child's disease, effects of medication on child's development, how to respond to an acute episode, available services, access to other parents and how to discuss with family/friends. Search strategy well defined. No formal description of search terms/phrases was used. No summary flow chart provided	Psychological, informational and social needs
Eatough et al (2013)	EU	To describe the personal experiences of parents and compare findings from a previous study	Juvenile Huntington's disease	Qualitative phenomenological study/in-depth semi-structured interviews/ participants recruited through clinical genetics and neurological services in Sweden, Netherlands, Italy and Poland/ n = 14 parents (13 mothers, one father)	Significant emotional guilt and anxiety reported by parents. Need to speak with other parents who can relate and understand. Parents describe their social life as limited and lacking spontaneity. Feel isolated/lonely. Lack of knowledge and understanding of health professionals. Parents need respite and time to themselves from caring duties. Participants gathered from a diverse cultural group. Fathers under sampled	Emotional, social, informational and physical needs
Fisher (2001)	UK	Identify the needs of parents within the literature	Chronically sick child	Review of the literature/articles published in English 1992−1997/n = 8 articles included in review	Parental need for definitive diagnosis for their child, it signifies a turning point; alleviates feelings of uncertainty. Need for information regarding child's illness. Parents often dissatisfied with level of information and/or difficult to access. Poorly conducted review. Search strategy not well defined. No formal description of the databases searched or search terms/ phrases used	Informational and emotional needs
Gallo et al (2008)	USA	Examine parental concerns relating to privacy and disclosure of information, insurance and health care costs,	Genetic conditions	Qualitative descriptive study/open-ended in-depth interviews/participants recruited from three community clinical sites/ n = 142 parents (86 families)	Parents reported financial strain, concerns about present and future health care costs associated with child. Difficulties balancing work/caring responsibilities and taking time off work. Employment/	Practical and social needs

		employment and schooling for their child			pursuing career was more challenging. Concerned about child's schooling, performance, psychosocial wellbeing, physical health, access to resources and school's ability to manage child's health needs. Study design and sample well defined. Limited probing undertaken during interviews. Parents of school-age children only	
Goble (2004)	USA	Examine the lived experiences of fathers	Chronic illness	Qualitative phenomenological study/ unstructured, informal open-ended interviews/lasting 15−30 min/participants recruited from a single paediatrician's office/$n = 5$ fathers	Greater financial strain reported on fathers due to wives quitting work to care for sick child and additional medical expenses/ travel costs. Fathers felt isolated, having a reduced social life, guilt associated with missed opportunities for quality time with partner. Biggest worry was for future health and wellbeing of child. Reasonably well conducted study. Small sample of educated, affluent Caucasian fathers.	Practical, social and emotional needs
Graungaard and Skov (2006)	DNK	To investigate parents reactions and coping strategies to receiving a diagnosis for their child	Severe physical or mental disability	Qualitative longitudinal study/semi-structured in-depth interviews/at two intervals; within 3 months disclosure of disability and two years post-disclosure/ participants recruited from three clinical departments/$n = 16$ parents (eight couples) interviewed separately	Parent's ability to cope is strongly influenced by the diagnostic experience. Feelings of uncertainty and fear for child's future. Need for information about child's condition. Amount and quality of information provided by health professionals influenced parent's level satisfaction with them. Study design and methods well defined. Unbalanced representation of interviewees (education level, socioeconomic status).	Emotional and informational needs
Hendriks et al (2000)	NLD	Determine the service needs of parents in Dutch therapeutic toddler classes	Motor and multiply disabled child	Quantitative longitudinal study/33-item questionnaire based on established surveys/participants approached at two separate time intervals; at commencement of therapeutic toddler class; and 10 months following/$n = 81$ families (81 mothers and 80 fathers)	Parental need for information concerning child's disability, future development and available services. Mothers needed more support in responding to others questions regarding their child and in accepting child's condition themselves. A well conducted study. Items from existing surveys were changed/adjusted to meet target setting. Additional items were added to final survey with no evidence of reliability and validity testing	Informational needs
Hummelinck and Pollock (2006)	UK	To explore the information needs of parents and their evaluation of information provided	Chronic illness	Qualitative study/semi-structured interviews/ $n = 20$ sets of parents	Parents viewed communication and information provision by health professionals to be inadequate and often confusing. Medical 'jargon' left parents	Informational needs

(*Continued*)

Table 1
Continued

Author (year)	Country	Study aim	Disease	Study design/methods/sample	Findings/critique of study	Domains of SCNF
		by health care professionals in relation to prescribed medicine-taking of their child			feeling overwhelmed. Information needs of parents varied depending on length of time from diagnosis. As parents gain confidence in caring for their sick child, their information needs change. At time of diagnosis, parents want to know everything, to feel involved in the management of their child. Reasonably well conducted study	
Kerr and McIntosh (2000)	SCO	Investigate the benefits of parent-to-parent support	Child born with congenital limb deficiency	Qualitative hermeneutic phenomenology study/in-depth interviews/lasted 40minutes-3hours/Participants recruited through four limb fitting centers, one plastic surgery unit and one national association for limb deficiencies/$n = 63$ families (34 couples, 29 mothers)	Emotional impacts of giving birth of a child with disability reported. Feelings of uncertainty for child not alleviated in the months following birth due to a lack of information. Parents felt health professionals had a lack of knowledge/ experience of disability. Parents felt isolated, made worse by parents minimizing public appearances with their child. Parents felt talking to other parents in a similar situation gave them a more positive outlook and ability to cope. Study design, sample and methods well discussed	Emotional, social and informational needs
Kerr et al (2004)	CAN	To provide a detailed review of the parental supportive care needs literature using the supportive care needs framework (SCNF)	Childhood cancer	Review of the literature/primarily research articles written in English and published between the years 1992−2002/$n = 49$ articles included in the review	Parents needed more information regarding child's cancer. Uncertainty about child's disease, treatment/management and future health. Emotional needs discussed in-depth. Parental need for social support. Positive relationships with health professionals aided parents coping. Need for financial support, managing home/ work responsibilities. Some parents sought support through religion, sought meaning for experiences/child's disease. Parents experienced a number of physical symptoms resulting from the impacts of child's disease. Seminal study investigating parental supportive care needs. Limited to parents of children with cancer. Appears to have limited search to fit literature within the existing SCN domains. No formal description of search terms used to identify relevant articles.	Informational, emotional, social, practical, spiritual and physical needs
Kerr et al (2007)	CAN	To identify the supportive care needs of parents	Childhood cancer	Mixed methods study/questionnaire based on modified version of an existing survey/	Parents struggled emotionally seeing their child suffer, fearful child's cancer would	Emotional, informational,

				telephone interviews 3 months post-survey/lasted 30–40 min/participants recruited through a pediatric oncology program within a tertiary care hospital and regional cancer centre/n = 15 parents (14 were mothers) completed questionnaire/ n = 3 parents were interviewed	spread and condition worsens. Parents felt a lack of control, powerless/useless, unable to help. Parents desired more written/verbal information, to be included in the care of their child and informed of all treatment options. Financial strain reported. Physical needs included sleep problems. Spiritual search for meaning within their experiences of having a child with cancer. A reasonably well conducted study. Small sample size. Fathers under-sampled. Details of reliability and validity testing of revised survey unclear.	practical, social, psychological, physical and spiritual needs
Kirk and Glendinning (2004)	UK	Explore the experiences of families caring for a child at home, and identify the perceived issues by professionals supporting parents regarding service provision, coordination of care	Technology-dependent child	Qualitative descriptive study/in-depth face-to-face interviews/participants recruited from three specialist children's hospitals/ n = 33 parents (23 mothers, 10 fathers) and n = 44 health professionals	Parents needed more support in planning/ coordinating multiagency services and professionals. Limited social life due to child's cumbersome medical equipment. Lack of spontaneity in life, need more respite from caring duties. Burden of care impacted on partner relationship. Parents needed more specialist knowledge/skills to properly care for their child, support from others who are properly trained, and health professionals to consider more carefully parents capacity to take on the complex technical care of their child. A well conducted study. Research design and methods well discussed. No limitations declared by authors.	Practical, social and informational needs
Lim et al (2012)	AUS	To identify cultural barriers and factors that exists to diagnosis for parents in China	Rett syndrome	Qualitative study/telephone interviews/lasted 0.5–1.5 h/participants recruited from a university hospital database/n = 14 families, all mothers	Parental need for early definitive diagnosis. Prolonged consultation and waiting times for parents added to feelings of stress, frustration and anxiety. Health professionals lack of knowledge of child's disease. Study design, sample and methods well defined. The term 'family' used to refer to mothers	Informational needs
McGrath (2001)	AUS	Discuss and compare issues of accessing support for parents across five treatment stages	Acute Lymphoblastic Leukemia	Qualitative phenomenological study/open-ended interviews/participants whose child was at the end of/to five weeks post-remission induction phase/participants were recruited from a hospital oncology ward/n = 12 families (12 mothers and 4 fathers)	Parents felt as time passed, provisional support became less; the longer the 'crisis' lasted, the less people offered them support. Need for practical and emotional support to help cope. Parents valued support from family, friends, health professionals and other parents. Parents felt that the level of support available to them correlates with their ability to cope.	Social, practical and emotional needs

(*Continued*)

Table 1
Continued

Author (year)	Country	Study aim	Disease	Study design/methods/sample	Findings/critique of study	Domains of SCNF
Nahalla and FitzGerald (2003)	LKA	Describe the experiences of parents with a child requiring regular hospitalization for blood transfusions	Thalassemia	Qualitative interpretive phenomenological study/in-depth interviews/lasting 0.5 h/ participants were recruited from a thalassaemia unit/n = 10 parents (7 mothers, 3 fathers)	Parents describe emotional needs around gaining access to the best health care services for their child. Financial difficulties associated with ongoing health care costs, and their capacity to work was impacted. Concern about neglecting/ limited time spent with siblings, and the impact chronic illness may have on them. Partner relationship impacted by child's illness. Small Sri Lankan sample	Emotional, practical and social needs
Neil-Urban and Jones (2002)	USA	Describe the experience of fathers and their coping strategies regarding their child'scondition	Childhood cancer	Qualitative phenomenological study/focus groups/lasted 2.5 h/participants recruited from a hospital-based program for treatment of childhood cancer/n = 10 fathers (7 fathers, 2 step-fathers, 1 grandfather)	Financial difficulties reported, feeling added pressure to provide, work productively and meet financial expectations. Fathers described feeling vulnerable, bottling up grief for prolonged periods, recalling their child's cancer as an emotionally traumatic experience. Persistent need to know why. Changed life priorities as a result of experience. The purpose of study not clearly discussed	Practical, emotional and spiritual needs
Palisano et al (2010)	USA	To identify perceived needs of parents, and whether the number and types of needs differ based on age and gross motor function levels of their child	Cerebral palsy	Quantitative cross-sectional study/ questionnaire based on established survey/ participants recruited from one of six major children's hospitals, and who were already part of a larger multisite study/ n = 501 parents (77.6% mothers)	Parents of children with greater requirements expressed more needs for information, community-based services and financial support. Parents in general needed more information regarding current and future services, planning, accessing supports/ activities for their child. Need for respite/ personal leisure time from caring duties and a skilled babysitter. Additional survey questions not tested for validity or reliability	Informational, social and physical needs
Pelentsov et al (2014)	AUS	To provide an in-depth account of the experiences and supportive care needs of parents	Ectodermal dysplasia	Mixed methods study/single focus group/ lasted 1−1.5 h/participants recruited from a national parent support group/n = 8 parents (6 mothers, 2 fathers/web-based questionnaire/50-items/n = 126 respondents (92% mothers)	Parental need for early definitive diagnosis; brought relief to parents. A diagnosis enabled them to better cope, manage child's rare disease, minimize risks, access support/other resources, answer questions and plan for the future. Health professional's lack of knowledge of disease and minimal support. Impact on partner relationships, parents felt they neglected the siblings. Only single focus group. Fathers under sampled	Emotional, social and informational needs

Speraw (2006)	USA	To examine the lived experiences of parents and caregivers seeking formal religious education for their children	Disabilities or special needs	Qualitative phenomenological study/open-ended face-to-face interviews/lasted 1−3 h/participants recruited via secular/religious-oriented publications, disability-related listservs and snowball techniques/n = 25 families (30 fathers, 14 mothers)	Parents felt alienated and devalued by the spiritual community for having a disabled child and the spiritual needs of their child were not recognized. Parents changed congregations in search of an accepting setting for their child. Parents felt socially disconnected from the spiritual community resulting in a crisis of faith. Details of recruitment could be made clearer. Parents in the study were all part of a faith group	Social and spiritual needs
Strehle and Middlemiss (2007)	UK	To explore the views and opinions of parents who experience an unexpected diagnosis in their child	4q-syndrome	Quantitative study/postal questionnaire/participants recruited via a national parent support group/n = 32 parents completed the survey	Parents reported strong emotions to receiving a formal diagnosis for their child. Ongoing feelings of uncertainty and insecurity about child's future. Need to speak with other parents in similar circumstances. Parents felt skeptical of health professionals, protective of their child. Small sample size. Details of how participants were recruited were not clearly discussed. Questions not shown to be valid or reliable. Respondent demographics not provided	Emotional and social needs
Trulsson and Klingberg (2003)	SWE	Describe the lived experiences of parents and their needs related to caring for their child	Orofacial problems	Qualitative grounded theory study/open-ended in-depth interviews/lasting 1 h/participants recruited at a specialized pediatric dental clinic/n = 14 parents (12 mothers and 2 fathers)	Parents describe caring responsibilities as never ending and felt pressure to remain functional. Describe strong emotions related to caring for their child. Health professionals lack of knowledge and understanding of child's condition. Need for recognition and validation by health professionals, to be listened too, respected and shown empathy/kindness	Emotional and informational needs
van Scheppingen et al (2008)	NLD	To identify and specify the problems experienced by parents caring for their child	Epidermolysis Bullosa	Qualitative study/semi-structured interviews/duration 1−1.5 h/participants recruited from a dermatology database at a university medical centre/n = 11 families (11 mothers, 5 fathers)	Parental fear and uncertainty regarding child's long-term health. Parents emotionally impacted seeing their child suffer; powerless to curtail it. Public humiliation and embarrassment; insensitivity by others. Mothers quit work to care for their child. Financial difficulties discussed. Impact on partner relationship, guilt about neglecting the other siblings. Challenges coordinating services/planning care discussed. Parents felt physically exhausted, need for respite/leisure time. Health professionals lack knowledge and skills of condition. Parental need for more information	Emotional, social, practical, physical and informational needs

(*Continued*)

Table 1
Continued

Author (year)	Country	Study aim	Disease	Study design/methods/sample	Findings/critique of study	Domains of SCNF
Weng et al (2012)	TWN	Explore the perspectives of primary caregivers of children with a rare genetic disorder	Russell-Silver syndrome	Qualitative exploratory study/in-depth face-to-face interviews/participants recruited in a leading medical centre/$n = 15$ caregivers (11 mothers, 2 fathers and 2 grandmothers)	Emotional feelings of uncertainty and worry about child's future. Health professionals lack of knowledge and expertise. Parental need for more information. Caring responsibility placed on parents, and whether can sustain long-term. Need for respite/personal time from caring duties. Limited social life. Reduced employment hours/leave place of employment	Emotional, informational, physical and social needs
Yiu and Twinn (2001)	CHN	Identify the needs of Hong Kong Chinese parents during hospitalization of their child for treatment following diagnosis	Childhood cancer	Qualitative descriptive study/semi-structured interviews at two time-points; following child's diagnosis and following six weeks hospitalization period/$n = 5$ parents (4 mothers and one father)	Strong emotions reported at time of diagnosis. Uncertainty and fear about child's future and how parents would cope long-term. Need for individualized, easily accessible information. Health professionals need to be sensitive when discussing diagnosis with parents. Need for support by family and health providers, to feel connected and gain confidence to care for their child. Small Chinese sample. Recruitment strategy of participants unclear	Emotional, informational and social needs
Zierhut and Bartels (2012)	USA	Discuss the lived experiences of parents coming to terms with their child's diagnosis, and factors that they found helpful or detrimental during the pre-diagnosis period	Fanconi Anemia	Qualitative descriptive study/semi-structured face-to-face and interviews/lasting 1–1.5 h/participants recruited from a university based FA specialist clinic/$n = 9$ parents (6 mothers, 3 fathers)	Parents reported the initial diagnosis period as a time of acute distress for them. Other strong emotions described. Ongoing uncertainty and concerns for sick child. Difficulties coping. Need for positive thinking strategies. Need for parental support groups/social supports. Financial difficulties impacted on time spent with child in hospital. A small, well conducted study	Emotional, practical and social needs

The second most common cited informational need concerned the level of knowledge and understanding by health professionals. Parents voiced frustration by the lack of knowledge and experience demonstrated by health professionals associated with their child's disease.[11,28,29,32–34,41] Poor communication and an inability to provide parents with a comprehensive prognosis for them and their child were a constant source of concern for parents.[28,33] Parents felt this lack of information hindered their ability to cope and manage their child's health when situations arose.[21,23,34] Equally, parents felt that they required considerably more specialist knowledge in order to care for their child.[31,32] In many cases, parents had to assume the role of 'expert'.[31,32,34] As a result, parents sourced relevant information themselves in order to answer their own questions, and allay concerns.[32] However, many parents found accessing information difficult.[12,34,37,40,42] They desired readily and easy to access information that was meaningful to them.[40] They also want educational resources/material that they can provide friends and family, and that they can use to answer questions about their child's disease.[11,23] Finally, parents valued receiving an early and definitive diagnosis for their child.[11,37,38,41]

Emotional needs

Parents described the initial diagnosis period as a highly emotional time for them. Strong emotions such as shock, distress, anger, fear, disbelief, denial and guilt were commonly felt by parents.[4,24,27,29,40,43] The most significant ongoing emotional needs expressed by parents were dealing with stress and guilt. Stress was manifested by anxiety, uncertainty, worry, fear, frustration, grief, powerlessness, shock, denial. Parents were, overwhelmed with sadness, feelings of vulnerability and suffered from anticipatory loss, blame, confusion, disbelief, dismay, helplessness, insecurity and a lack of control.

Most commonly, the emotional needs of parents were associated with their child's health needs, the severity of the child's disease and parents' caring responsibilities. On a daily basis parents felt overwhelmed regarding their sick child, and whether they had the capacity to cope long-term with the day-to-day demands and caring responsibilities.[13,22–24,29,32,33,35,43,44] Some parents lived in fear and helplessness that their child's disease would spread or that their child's disease would worsen over time.[13]

Overall, parents expressed uncertainty and insecurity for the future health and wellbeing of their sick child.[12,22,27,30,33,38]

Parents felt their managing and coping abilities were largely influenced by their diagnostic experiences.[38] For some parents, receiving a diagnosis in their child was one of relief. It signified an emotional turning point for them from one of uncertainty.[11,37] In part, parents did not feel that health professionals spent adequate time addressing their emotional needs, and would have valued a higher level of knowledge and support from them.[21,27,28,41]

Practical needs

Financial concerns were the main practical need of parents.[4,12,13,23,30,35,36,43,45] Parents described financial matters as a major source of stress and distress for them.[13,23,43] Many parents reported significant financial hardship associated with their child's disease and ongoing care requirements.[4,35] Due to the daily care requirements of their child, mothers in particular had to reduce their working hours or quit full-time employment in order to stay home and care for their sick child. This placed an added burden on fathers to remain the sole provider for the family with additional income required to cover medical expenses.[30,45] Parents voiced concerns regarding present and future health care costs associated with their child.[36] Parents felt that in order for others to care for their child in their absence, a certain level of training of the carer was required so that parents could feel that their child was safe and in good care.[31] Planning and coordinating care and services to meet their child's unique care needs was also a constant source of stress and concern for parents.[31,32] Parents needed respite or time away. With the everyday caring demands and stresses caring for their sick child, parents most desired more respite or personal leisure time free from their caring responsibilities.[26,28,32]

Physical needs

Caring for their child often left parents feeling physically and mentally exhausted.[32] Other physical symptoms experienced by parents related to the impacts of caring for their sick child include sleep disturbance, fatigue, loss of appetite, weight loss, headaches, dizziness and frequent colds.[12,13,32] Some parents themselves had the rare disease, either fully or as carriers, and suffered from physical problems associated with it.[11]

Spiritual and psychological needs

Authors cited spiritual and psychological needs much less in the studies compared to the other domains within the SCNF. The spiritual needs of parents involved them searching for meaning within their experiences of having a child diagnosed with a rare disease.[12,13,45] Through their search to find meaning, parents gained new insights which helped change their views on what they considered important.[45] For some parents, not feeling connected or 'belonging' to a church community because of their child's disease resulted in them having a crisis of their faith.[25] The psychological needs of parents were often described by parents as them feeling useless, powerless and helpless in their care for their child with a rare disease.[13,22,38] Despite feeling emotionally distressed and in a state of shock, few parents were offered psychological support or counseling from mental health

Table 2
Summary of themes identified

Type of need		*n*	%
Social needs (21 papers)	• Balancing work and family, employment, work hours	3	14.3
	• Collaboration, partnerships with health professionals	3	14.3
	• Dealing with public situations/societal expectations	3	14.3
	• Difficulties accessing support	3	14.3
	• Feeling isolated, loneliness	7	33.3
	• Need more support from health professionals, family and friends	9	42.8
	• Need to speak with other parents	8	38.1
	• Relationships; partner and siblings	5	23.8
	• Social life, need for spontaneity	6	28.6
Informational needs (19 papers)	• Having information that is readily available and easy to access	3	15.8
	• How to discuss child's condition/answer questions	1	5.3
	• How to respond to an acute episode of illness in their child	1	5.3
	• Identifying own informational needs	1	5.3
	• Level of knowledge/understanding by health care professionals	8	42.1
	• Need for a definitive diagnosis	4	21.0
	• Need for education material to provide family and friends	2	10.5
	• Need for information regarding their child's disease	13	68.4
	• Having to assume the role of 'expert'	3	15.8
	• Receive information on child's future health, development and services	4	21.0
Emotional needs (18 papers)	• Anger	3	16.7
	• Anticipatory loss, blame, confusion, disbelief, dismay, helplessness	1	5.5
	• Anxiety, uncertainty, worry	7	38.9
	• Denial, overwhelmed, sadness, vulnerable	2	11.1
	• Fear, frustration, grief, powerlessness, shock	4	22.2
	• Guilt	10	55.5
	• Stress	12	66.7
	Emotions related to:		
	• Around time of diagnosis	8	44.4
	• Child's future health, health care needs and available services	10	55.5
	• Child's needs, severity of child's disease, own caring responsibilities	13	72.2
	• Finances; employment/work	7	38.9
	• Level of knowledge and support by health professionals	3	16.7
	• Relationships, partner/siblings	2	11.1
Practical needs (10 papers)	• Childcare, reliance on others to care for child	1	10.0
	• Finances; work/employment matters	7	70.0
	• Legal, health care services and entitlements	1	10.0
	• Need for respite, time off from caring responsibilities	4	40.0
	• Organization and coordination of care for child	1	10.0
Physical needs (6 papers)	• Physical symptoms experienced e.g. sleeping problems, fatigue	3	50.0
Spiritual needs (4 papers)	• Crisis of faith, challenging own spirituality	1	25.0
	• Search for meaning, makes sense of the situation, gain new insights	3	75.0
Psychological needs (3 papers)	• Self-worth	1	33.3
	• Psychological stress and distress	1	33.3

professionals, counselors or peer support groups, particularly around the time of diagnosis; when need was greatest.[4]

Table 2 summarises the findings with respect to reported needs from the 29 papers reviewed in this scoping study. Clearly, social, informational and emotional needs dominated in the published literature, with the most common being the need for information about their child's disease, stress, guilt and uncertainty about their child's disease and future health, and the need for more support.

Discussion

We have found the use of the SCNF for parents of a child with a rare disease was challenging. In particular, we encountered difficulties in the classification of parent needs cited within the literature across the seven domains of the SCNF. Some needs we found fit quite well and some did not; while others, we felt fit across more than one domain. Further, some reported needs, such as isolation and loneliness, were simply not part of the SCNF.

In studies by Kerr et al (2004; 2007), they encountered difficulties categorizing the needs of parents of children with cancer using the SCNF. If the SCNF is not entirely suitable for parents of a child with a rare disease, how could it be modified to make it a better fit? Is there a need for one or more new domains, additional categories within each domain, or the removal of non-relevant categories or domains? According to Kerr et al,[12] a prospective assessment of the needs of parents from their perspective would be the

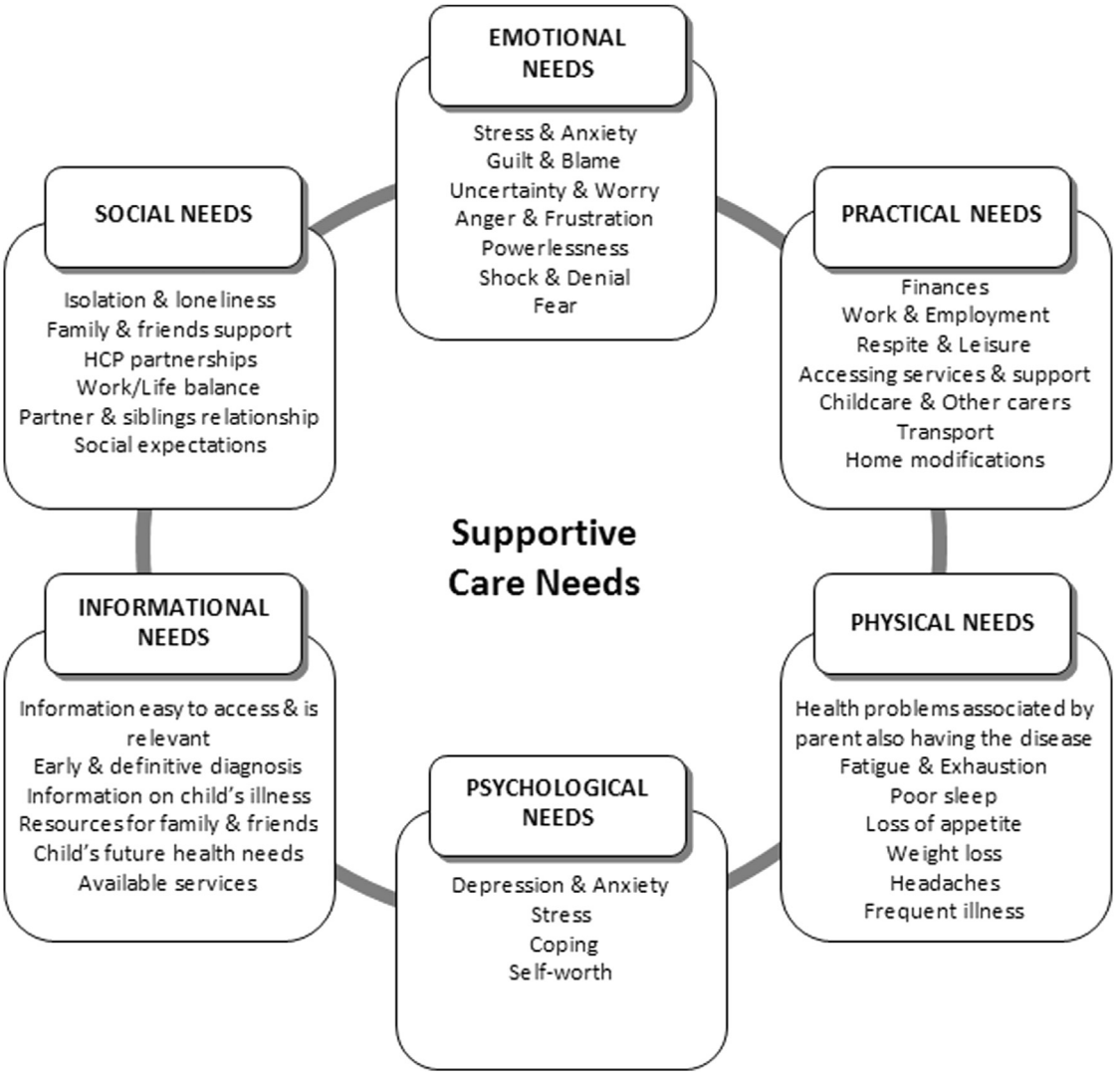


Fig. 3. Proposed parent supportive care needs framework.

next logical step forward in gaining an understanding of their specific and unique needs. Yet to date, there has been no study which has attempted to examine the entire constellation of supportive care needs of parents.[13] Based on findings of this scoping review, we suggest (see Fig. 3) a modification to the SCNF that we consider more appropriate. This is a preliminary suggestion for a conceptual framework, and there clearly is a need for much more research in this area, in particular, empirical justification for the proposed framework. We agree with Kerr et al,[12] who pointed out that if needs are not well defined they cannot then be satisfactorily met in the provision of care to patients and their families.

From our own research, we have found a number of items that we feel should also be added to the framework. Such as, parents themselves having the rare disease and it causing physical symptoms in them, and parents need for an early definitive diagnosis for their child.[11]

The authors of the SCNF acknowledged that a person impacted by a life-threatening illness will experience more than just physical changes; they will also be impacted by psychological, social, practical and spiritual consequences.[46] Yet, each of the domains within the SCNF appears siloed. The SCNF does not seem to describe or imply any suggestion of causality. Yet, it is apparent that some needs are likely to cause others. For example, some practical needs of parents (e.g. financial needs) are likely to lead on to emotional needs, which if not resolved, can lead to psychological distress. In future iterations of the new framework we will consider potential causal pathways. Although it is clear that different rare diseases may impact on the affected children in many different and often unique ways, it is also clear that many of the needs of the parents are universal. For example, having a child with a rare disease is almost certainly likely to impose a financial burden on the parents of that child. We therefore feel it is sensible

to study the commonality of supportive care needs across affected parents, something which has not been done previously across the full spectrum of rare diseases.

Limitations of the review

This review followed the seminal scoping study framework proposed by Arksey and O'Malley.[18] It consisted of an iterative process whereby the authors have engaged with each of the five stages of the scoping review process in a reflexive manner, and where necessary, repeating steps to ensure that the review of the literature in this area has been comprehensive.[18] Despite this, there are some limitations associated with the conduct of this scoping review that need to be acknowledged. The primary limitation was the search strategy was limited to English-language studies only, and there may well be other literature equally relevant to the area of parental supportive care needs in rare diseases that we simply do not know exists because it was published in another language. The majority of the studies reviewed had mother's voices more prominent – despite the title of the paper invariably using the word "parent". Less is known about what fathers are saying, and we do not assume that parents supportive care needs are homogenous.

Implications for further research and practice

The paucity of studies in this review reflects a lack of focus on parents (particularly fathers) needs caring for a child diagnosed with a rare disease. This lack of research is surprising, given the global impact of rare diseases, and the fact that the burden and caring responsibilities of children suffering from chronic diseases typically falls on the parents.[47] The results of this review emphasize the lack of evidence and highlights a need for further research on this important area. In particular, the development of a revised parental SCNF that looks specifically at the supportive care needs of parents of a child with a rare disease is warranted.

Conclusion

The seven domains of supportive care needs outlined in the SCNF are cited within the literature to varying degrees, with social, informational and emotional needs being the most common. We conclude that using the SCNF for parents of a child with a rare disease is challenging. There are difficulties in the classification of parent needs cited within the literature across the seven domains outlined in the SCNF. Some needs we found fit quite well and some did not, while others, we felt fit across more than one domain. Furthermore, we identified a number of items which we feel should also be added to the framework. It is evident from this review that the SCNF is not entirely suitable for parents of a child with a rare disease. There is an urgent need for more research to be conducted, particularly the development of a revised parental SCNF which looks specifically at the supportive care needs of parents of a child with a rare disease and potential causal pathways. Furthermore, this review highlights a need for more research to be conducted looking at the entire constellation of supportive care needs of parents with a child with a rare disease, irrespective of which disease their child has, in order to satisfactorily meet their needs in the provision of care of these families living with rare diseases.

Key messages

- Parents caring for a child diagnosed with a rare disease report similar unmet needs; and irrespective of their child's disease and sequelae, parents' needs are homogenous.
- This review is the first to provide a detailed synopsis of parental supportive care needs literature published through 2014.
- There is a need for more research to be conducted looking at the entire constellation of supportive care needs of parents with a child with a rare disease; particularly the development of a revised parental SCNF which looks specifically at the supportive care needs of parents of a child with a rare disease and potential causal pathways.

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