Understanding burnout among parents of children with complex care needs: A scoping review informed by a stakeholder consultation

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Abstract

Background:

The concept of parental burnout has been proposed to be helpful for describing and understanding the impact of parenting children with complex care needs (CCN). The objective of this scoping review was to investigate the aims, methods, and results of research on burnout among parents of children with CCN, and to inform the interpretation as well as implications for research and practice through a stakeholder consultation.

Methods:

The scoping review was guided by the JBI Reviewers' Manual for scoping reviews. Primary studies on burnout among parents of children with CCN were identified through a comprehensive search of peer reviewed and grey literature. Metadata and findings were extracted and compiled for further synthesis and interpretation, guided by a stakeholder consultation, involving parents of children with CCN ($n = 6$), healthcare professionals ($n = 3$), and researchers ($n = 3$). The stakeholder consultation was structured according to a Delphi consultation method.

Results:

A total of 57 articles were eligible for inclusion. Prevalence of burnout among parents of children with CCN varied between 20 and 77%. Parents of children with CCN reported on average higher burnout scores than parents of children without CCN. While all studies focused on parents with children with CCN, very few studies specifically focused on burnout within the context of parenting a child with CCN. Stakeholders endorsed the importance of studies into the multifactorial determination of burnout in the context of having children with CCN.

Conclusion:

The concept of parental burnout highlights the extremes of stress and burden experienced by parents of children with CCN. By focusing more on contextualized definitions of parental burnout, research may unravel the complex interplay between personal and contextual factors pertaining to risk and resilience.

Keywords: parental burnout, caregiver burnout, children with complex care needs, children with special needs.

1. Introduction

Parenting is rewarding but challenging. This is particularly true among parents of children with complex care needs (CCN), defined as “...those who have or are at increased risk of chronic physical, developmental, behavioral, or emotional conditions and who also require health and related services of a type or amount beyond that required by children in general” (McPherson et al., 1998, p. 138). Parents of children with CCN face unique demands such as dealing with uncertainty about their child’s condition, performing complex in-home caregiving tasks, and adapting their routine to fit the needs of the child (Power, Dahlquist, & Pinder, 2019). Studies have shown increased levels of parenting stress (Cousino & Hazen, 2013; Golfenshtein, Srulovici, & Medoff-Cooper, 2015) and higher levels of anxiety and depression among parents of children of CCN compared to parents of typically developing children (Barreto et al., 2020; van Oers et al., 2014). The concept of ‘parental burnout’ has been introduced to capture how prolonged exposure to parenting-related risks may have severe long-term psychosocial effects. Parents have been advocating for research attention towards the risk of prolonged parenting-related strain, parental burnout, and potential mitigating factors (Bjornstad et al., 2019; McIntyre, Novak, & Cusick, 2010; Vereniging van mensen met een lichamelijke handicap, 2018).

Parental burnout has been conceptualized as a manifestation of exhaustion due to long-term emotional strain and insufficient resources for coping (Hubert & Aujoulat, 2018), although other definitions have been proposed as well. It has been characterised along the interrelated dimensions of (1) emotional exhaustion related to one’s parental role, (2) emotional distancing from one’s child, (3) contrast with previous parental self, and (4) feelings of being fed up with one’s parental role (Roskam, Brianda, & Mikolajczak, 2018). Emotional exhaustion refers to the feeling of being immensely drained, and no longer being able to carry on in one’s parental role. Such feelings may result in a detached stance towards the child, limiting the interactions to functional aspects at the expense of emotional investment. Furthermore, parents may think less of themselves as parents and lose the enjoyment of being there for their child (Hubert & Aujoulat, 2018; Roskam, Brianda, & Mikolajczak, 2018).

In the general population, parental burnout has been described in relation to negative outcomes among parents themselves, for example, escape and suicidal ideation, somatic complaints and domestic conflicts (see for example Mikolajczak, Brianda, Avalosse, & Roskam, 2018; Sarrionandia-Pena, 2019) and for their children (Hubert & Aujoulat, 2018; Mikolajczak et al., 2018). A recent study in 42 countries showed that the prevalence of burnout among parents from the general population globally ranged between 0% and 9% (Roskam et al., 2021). Given the potentially detrimental impact of parental burnout and

considering the additional challenges and demands that parents of children with CCN face, families with children with CCN may be particularly at risk. Moreover, the unique context of parents of children with CCN may translate into unique features of parental burnout as well as a different set of associated factors. Sánchez-Rodríguez, Perier, Callahan, and Séjourné (2019) conducted a systematic review on parental burnout in the general population, providing an overview of sociodemographic- and personality characteristics of parents and children, effectiveness of interventions aimed at parental burnout, as well as clues to explaining the phenomenon of parental burnout. Their study showed a surge in the research interest towards parental burnout and approximately 60% of all the identified studies concerned parents of children with CCN. Given the recent rise in research interest on parental burnout and a high amount of research focussing on parents of children with CCN, summarizing the scope of studies focussing on burnout among these parents would be pertinent.

Scoping reviews of emerging research fields help to more narrowly focus questions and research designs into particularly fruitful and relevant directions (Munn et al., 2018). For weighing the social validity of the research and the relevance of the remaining questions and gaps, a stakeholder consultation may be included as part of the method for conducting such a review (Levac, Colquhoun, & O'Brian, 2010). Scoping reviews may also incorporate a stakeholder consultation by linking the knowledge from literature with the practice and the daily lives of those that the topic concerns (Arksey & O'Malley, 2005; Levac et al., 2010). However, stakeholder consultations are not often integrated in scoping reviews (Pham et al., 2014), and the experiences and perspectives of parents, healthcare professionals and researchers have yet to be included in reviewing research on parental burnout.

The objective of this scoping review was to describe scientific progress in describing and understanding burnout among parents of children with CCN and identify important remaining gaps. Specifically, the following research questions were investigated: (1) how is parental burnout conceptualized, (2) what differences have been described compared to parents of children without CCN on burnout scores and/or on the dimensions of burnout, (3) what is known about prevalence, and (4) what associated factors have been studied that relate to burnout. Answers to these questions, conclusions, and recommendations were formulated by taking into account the perspectives of parents, healthcare professionals, and researchers as important stakeholders in this research field.

2. Methods

2.1. Design

The scoping review implemented the JBI Reviewers' Manual (Peters et al., 2020). In addition, a stakeholder consultation was structured according to a Delphi consultation

method. Hence, this study took a participatory research approach. The objectives and methods of this scoping review were described in a protocol registered on Open Science Framework, and can be accessed via

https://osf.io/afs8x/?view_only=b8142a3d75e443dab64d10d150e9f943.

Scoping review phase

2.2. Identifying relevant studies

The literature search was performed in collaboration with a university librarian (MM). On July 3, 2020 the following academic databases were searched from day of inception: Medline (PubMed), Embase.com, Web of Science, Scopus, PsycINFO (Ebsco), CINAHL (Ebsco) and ERIC (Ebsco). Google Scholar and sources for grey literature (opengrey.eu, lens.org, oaister.worldcat.org, science.gov, base-search.net, and diva-portal.org) were searched in August 2020. The grey literature was searched with the aim to complement the peer-reviewed literature, not to be exhaustive. The search strategies combined the key words “parents” and “burnout” using their respective thesaurus terms and synonyms for free-text searching. No limitations were applied, except for comments, editorials and letters. The search strings for each academic database are provided in Supplement 1. To retrieve relevant records from grey sources the phrase “parental burnout” was used.

2.3. Selecting the studies

All records retrieved from the academic databases were imported into EndnoteX9, and duplicates were removed (Bramer, Giustini, de Jonge, Holland, & Bekhuis, 2016). The remaining records were uploaded in Rayyan QCRI (Ouzzani, Hammady, Fedorowicz, & Elmagarmid, 2016) for independent screening. For Google Scholar and the grey literature sources, articles were screened in the databases. To ensure that the eligibility criteria were sufficiently clear to both reviewers (NP and KM), first a random selection (10%) of the retrieved records was independently screened on titles and abstracts; discrepancies were discussed between NP, KM, and CS. Subsequently, all records were screened against the eligibility criteria by NP and KM. CS was consulted in the event of any discrepancies. This resulted in an inter-rater reliability of 90%. The full text screening was done by NP, who consulted KM in case of doubt. Lastly, the references of the included studies were searched manually by NP to detect additional relevant studies. In the event that studies were deemed eligible but no access to full text articles was available, NP or KM contacted the first author to request the full-text article.

Studies were eligible for inclusion when burnout among parents of children with CCN was the focus of the study and was measured or observed as a variable in the study. Articles focusing on parents of typically developing children and burnout in another context than

parenting (i.e. occupational context) were excluded. Furthermore, primary studies employing a qualitative, quantitative or mixed methods approach and case studies were eligible. We included peer reviewed publications (academic journal and book series) and grey literature (PhD dissertations and book chapters). To be eligible, full texts had to be available and published in languages accessible to the authors (English, Dutch, Swedish, Finnish, French, German).

2.4. Charting, collating and summarizing the data

Data were extracted using Google Forms. The extraction form was pilot-tested several times and adjusted accordingly. NP systematically extracted the data, KM and CS were consulted when necessary. Supplement 2 contains an overview of some of the items extracted. The extracted data were summarized in tables or thematically analyzed per research question, by using an open coding strategy. This was done by NP and one project group member assigned per research question. Prior to the stakeholder consultation, the extracted data was summarized by NP, KM and AW.

2.5. Stakeholder consultation

A stakeholder consultation was applied to inform the interpretation of the extracted data and to provide multiple perspectives in the interpretation and implications for practice and future research. The stakeholder consultation consisted of a convenience and purposefully chosen sample of parents of children with CCN ($n = 6$), professionals working with parents and/or families with children with CCN ($n = 3$), and researchers familiar with research on parental burnout ($n = 3$). We purposefully chose stakeholders who we expected to be reflecting on burnout among parents of children with CCN. Some of the stakeholders had a double role: two parents and two researchers also worked as healthcare professionals. For all participants the following exclusion criteria applied: (1) having received any psychiatric treatment in the past six months, related to burnout; (2) waiting for psychiatric treatment related to burnout; (3) being suicidal.

Stakeholders were contacted via email with an information letter and informed consent form. Those eligible and consenting to participate were encouraged to partake in a three-step process: (1) an individual interview, (2) two online surveys, and (3) a focus group session. However, stakeholders were spread over the different rounds and not all participated in each round (Figure 1). After each step, the qualitative data were reviewed by A and B and discussed with the project team to guide the next step. The individual interviews were facilitated by two project members and five project members were present during the focus group. A topic list was used to guide the interview and the focus group. The data from

the online surveys were collected through Survalyzer. For the topic lists and questionnaires see Supplement 3.

2.5.1. Analysis

Based on the objective of the stakeholder consultation, we analyzed the qualitative data according to the following questions: (1) do stakeholders relate with the results from the literature?, (2) what do stakeholders emphasize?, and (3) what aspects do stakeholders bring up that are not mentioned in the literature? The data were analyzed using a thematic analysis guided by the research questions of this review. The three questions used to analyze the stakeholder consultation also formed the initial coding categories. After assigning the initial coding categories, the categories were further examined to detect potential subcategories. A conducted the analysis and project members were assigned to critically examine the results per research question. After the final step of the stakeholder consultation, a draft version of the results was presented to all stakeholders for a member check.

2.5.2. Reflexivity

The project group consisted of two parents of children with CCN who initiated the project, a professional working with families of children with CCN, and four researchers. Furthermore, two additional parents of children with CCN were involved in the early stages of the study to provide input and determine the research direction. The participatory approach led to an emphasis on the involvement of stakeholders at every step of the research and on implications for practice.

Figure 1. The process of the stakeholder consultation

3. Results

3.1. Main study characteristics

Academic databases initially yielded 3,629 records. After deduplication, 1,843 records remained for screening of titles and abstracts on eligibility. Of these, 201 articles were selected for full text screening. This resulted in 50 studies meeting the inclusion criteria. One additional study was identified from backward snowballing and 6 studies were retrieved from additional databases. This resulted in the final inclusion of 57 studies. The full selection process is given in the PRISMA flowchart (Figure 2).

Figure 2. PRISMA flow diagram for study selection

The characteristics of the included studies are provided in Supplement 4. Publication dates ranged from 1985-2020 (median year was 2017). Forty-three articles used a quantitative research design, 11 a qualitative research design, and three a mixed methods design. Among the quantitative studies, most studies employed a cross sectional design ($k = 32$), followed by experimental design ($k = 10$), and longitudinal design ($k = 1$). A large number of the studies were conducted in Europe ($k = 22$), followed by the Middle East ($k = 19$), and North America ($k = 9$). The rest of the studies were spread over the world (Asia $k = 3$, South America $k = 2$, Africa $k = 1$, Australia $k = 1$). The most common type of respondent concerned parents irrespective of gender ($k = 31$), followed by mothers ($k = 17$), and primary caregivers ($k = 4$). Three studies concerned clinicians, of which one in combination with parents, and one study parents and teachers. Only one study focused on fathers alone. Regarding the children, 36 of the studies focused on one or two specific conditions of the child, while the rest investigated cross-syndromal CCN. Autism ($k = 8$) and diabetes ($k = 7$) were often studied. However, there was a large overlap between the conditions studied. Twenty-four studies focused on parents of children ≤ 18 years, while eight studies reported on parents of children also > 18 years. However, among eight studies the age range of the children was not mentioned and 17 studies did not report any information regarding age.

3.2. Conceptualization of burnout

From a total of 57 studies, 46 studies used a predetermined definition of burnout. Three sets were identified. First, burnout was often defined according to symptoms, such as 'exhaustion' of emotional, physical and/or cognitive nature. Second, others described burnout according to its onset factors, such as 'chronic stress'. Third, at times burnout was described according to its pathogenesis, such as when demands exceeded the coping and motivational resources. The remaining 11 studies all had a qualitative design and explored the conceptualization of burnout or had a description or a definition of burnout in the results or the discussion section of the article. These studies also added to the description of unique caregiving tasks of parents of children with CCN and the unique contextual factors, such as having to advocate for adaptations in care and education and dealings with health care providers, professionals, and responses from family and friends.

Studies that were based on predetermined definitions of burnout addressed parental demands across domains, such as work and career, the partner relationship, and caregiving (i.e. as a consequence directly related to having a child with CCN). As such, some studies used burnout in a broad sense, even within the context of parenting a child with CCN. When burnout was framed as pertaining to a specific context, terms such as, "parental burnout", "diabetes burnout", "caregiver burnout", and "couples burnout" were mentioned. This was reflected in the way in which burnout was measured. Studies often used inventories which

could not be traced as distinctly measuring burnout in the context of parenting or caregiving. Forty-seven studies used one questionnaire to measure burnout, one study applied two questionnaires to measure burnout. The most common questionnaire used was the Maslach Burnout Inventory (MBI) or a derivative of it ($k = 19$), followed by the Shirom-Melamed Burnout Questionnaire (SMBQ) ($k = 17$), and 10 other questionnaires which were applied ≤ 3 times. The MBI and SMBQ are not created to specifically measure burnout within a parenting context, but rather for an occupational context (Maslach, Jackson, & Leiter, 1997; Melamed et al., 1999). Common dimensions used to measure burnout were for example, exhaustion or being fatigued, distancing, lack of accomplishment, and tension. Well-validated questionnaires to measure burnout in the parenting context are the Parental Burnout Inventory (PBI; Roskam et al., 2017), which was derived from the MBI, and the Parental Burnout Assessment (PBA; Roskam et al., 2018), which was based on interviews with five parents suffering from exhaustion.

Stakeholders focused during the consultation on a fitting label for burnout among parents of children with CCN. Stakeholders emphasized that burnout stemmed from parental caregiving, distinct from the caregiving that professionals provide to the child. Therefore, the stakeholders proposed that a term combining both 'caregiving' and 'parenting' would be most appropriate, for example 'parenting caregiver burnout'. Yet, the term 'caregiving' did not have the same meaning for all stakeholders; some felt that it was associated with other types of caregiving, such as caring for an elderly parent or caregiving related to an occupational context. Also, caregiving failed to cover the link between children's dependence and life-long commitment as a parent. Consequently, no term received general agreement among stakeholders.

3.3. Differences between parents of children with and without CCN

Nine quantitative studies investigated the differences in burnout scores among parents of children with CCN compared to parents without children with CCN (Table 1). Among these studies, four calculated an overall burnout score and seven studies investigated the differences according to the different dimensions of burnout. All nine studies showed that on average burnout scores among parents of children with CCN were higher compared to parents without children with CCN. This was the case for studies that investigated burnout symptoms in general (measured with the SMBQ or MBI), but also for the study by Grain and Zech (2018) which was the only study measuring burnout symptoms in the parenting domain (PBI). Also, subscale scores were higher for parents of children with CCN, specifically on the dimensions of exhaustion and loss of efficacy, and somewhat less for

emotional distancing. Mean scores could not be compared across studies due to the heterogeneity of conceptualization and operationalization.

Table 1. Differences in burnout scores between parents of children with CCN and parents of children without CCN

Author (year)	Country of publication	Diagnosis/ condition of the child	Inventory used to measure burnout (reference), number of items, number of subscales, (number of items per subscale), Likert scale, mean or sum score, interpretation	Sample size among parents of children with CCN (n) (divided according to condition of the child/ type of parent)	Burnout scores (total and/or per dimension) among parents of children with CCN divided according to dimension, type of parent,	Sample size among parents of typically developing children (n)	Burnout scores (total and/or per dimension) among parents without children with CCN
Gérain and Zech (2018)	Belgium	Children with special needs	PBI (Roskam et al., 2017), 22 items, 3 subscales (8-8-6), 0-6 Likert scale, sum scores, higher scores indicate more burnout symptoms, except for PA which is reversed	n = 175 parents	Parents of children with at least one special need: EE mean: 17.7 * ED mean: 7.9 * LPA mean: 8.4 *	n = 725 parents	EE mean: 12.8 ED mean: 5.4 LPA mean: 7.0
Weiss (2002)	USA	Autism and mental retardation	MBI (Maslach et al., 1997), 22 items, 3 subscales (8-8-6), 0-6 Likert scale, sum scores, higher scores indicate more burnout symptoms, unclear if PA is reversed	n = 40 mothers of children with autism n = 40 mothers of children with a mental retardation	Mothers of children with autism: EE mean: 31.3 * DP mean: 9.5 * PA mean: 22.0 * Mothers of children with a mental retardation: EE mean: 27.6 * DP mean: 6.3 * PA mean: 20.5 *	n = 40 mothers	EE mean: 23.8 DP mean: 5.2 PA mean: 8.9
Basaran, Kardavut, Uneri, Balbaloglu, and Atasoy (2013)	Turkey	Cerebral palsy (CP)	MBI Turkish version (Duygun & Sezgin, 2003), 22 items, 2 subscales (14-8), 0-4 Likert scale, sum scores, higher scores indicate more burnout symptoms, except for PA which is reversed	n = 143 parents	EE mean: 14.0 * DP mean: 3.7 PA mean: 22.0 *	n = 60 parents	EE mean: 10.6 DP mean: 2.6 PA mean: 26.2
Senses Dinc, Cop, Tos, Sari, and Senel (2019)	Turkey	Down syndrome (DS)	MBI Turkish version (Duygun & Sezgin, 2003), 22 items, 2 subscales (14-8), 0-4 Likert scale, sum scores, higher scores indicate more burnout symptoms	n = 75 mothers	EE median: 30 PA median: 21*	n = 54 mothers	EE median: 29 PA median: 28

Kurtoğlu and ÖZÇİrpici (2019)	Turkey	Intellectual disability and/or physical disability	MBI Turkish version (Duygun & Sezgin, 2003), 21 items, 2 subscales (13-8), 0-4 Likert scale, sum scores, higher scores indicate more burnout symptoms, except for PA which is reversed	n = 97 mothers	Mothers of children with at least one intellectual disability and/or physical disability EE mean: 13.3 * PA mean: 27.8	n = 97 mothers	EE mean: 7.4 PA mean: 26.9
Dürüst (2018)	Cyprus	Children with special needs	BS (Pines, Aronson, & Kafry, 1981), Turkish version (Çapri, 2006), 21 items, 3 dimensions (7-7-7), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms	n = 120 parents	Mean: 3.6	n = 120 parents	Mean: 2.9
Lindahl Norberg (2007)	Sweden	Malignant brain tumour survivors	SMBQ (Grossi, Perski, Evengård, Blomkvist, & Orth-Gomér, 2003; Melamed, Kushnir, & Shirom, 1992; Melamed et al., 1999), 22 items, 4 subscales (8-4-4-6), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms	n = 24 mothers n = 20 fathers	Total mean score: 3.9 * mothers 3.0 fathers Mean burnout score per dimension among mothers: Exhaustion/ fatigue: 4.0 * Listlessness: 4.1 Tension: 3.9 Cognitive difficulties: 3.8 * Mean burnout score per dimension among fathers: Exhaustion/ fatigued: 3.0 Listlessness: 3.6 Tension: 3.2 Cognitive difficulties: 2.6	n = 152 mothers n = 107 fathers	Total mean score: 3.3 mothers 2.8 fathers Mean burnout score per dimension among mothers: Exhaustion/ fatigue: 3.2 Listlessness: 3.5 Tension: 3.4 Cognitive difficulties: 3.0 Mean burnout score per dimension among fathers: Exhaustion/ fatigue: 2.8 Listlessness: 3.2 Tension: 3.0 Cognitive difficulties: 2.5
Lindahl Norberg, Mellgren, Winiarski, and Forinder (2014)	Sweden	Children who had undergone hematopoietic stem cell transplantation (HSCT)	SMBQ (Grossi et al., 2003; Melamed et al., 1992; Melamed et al., 1999), 22 items, 4 subscales (8-4-4-6), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms	n = 159 parents	Total mean score: 3.4 mothers 2.9 fathers	n = 237 parents	n.s.

Lindström, Åman, and Norberg (2010)	Sweden	Type 1 Diabetes Mellitus (T1DM), Inflammatory Bowel Disease (IBD)	SMBQ (Grossi et al., 2003; Melamed et al., 1992), 22 items, 4 subscales (8-4-4-6), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms	<p>n = 251 parents of children with T1DM</p> <p>n = 142 mothers of children with T1DM</p> <p>n = 109 fathers of children with T1DM</p> <p>n = 38 parents of children with IBD</p> <p>n = 21 mothers of children with IBD</p> <p>n = 17 fathers of children with IBD</p>	<p>Total mean burnout score for mothers: 3.6 * DM 3.7 IBD</p> <p>Total mean burnout score for fathers: 3.2 DM 2.8 IBD</p> <p>Mean burnout scores for mothers, according to dimension (SMBQ) and condition of the child:** Exhaustion/ fatigue: 3.6 DM 3.6 IBD Listlessness: 4.0 DM 4.3 IBD Tension: 3.8 DM Cognitive difficulties: 3.4 DM 3.3 IBD</p> <p>Mean burnout score for fathers, according to dimension (SMBQ) and condition of the child: ** Tension: 3.6 DM</p> <p>Mean burnout scores for mothers, according to dimension (KES) and condition of the child:** Memory: 2.7 DM 2.9 IBD Physical symptoms: 2.1 DM 2.0 IBD Sleep disturbance / fatigue: 2.6 DM 2.7 IBD Emotions: 2.8 DM</p>	n = 124 parents	<p>Total mean burnout score for mothers: 3.0</p> <p>Total mean burnout score for fathers: 2.9</p> <p>Mean burnout score for mothers, according to dimension (SMBQ): Exhaustion/ fatigue: 2.9 Listlessness: 3.3 Tension: 3.2 Cognitive difficulties: 2.8</p> <p>Mean burnout score for fathers, according to dimension (SMBQ): Tension: 3.1</p> <p>Mean burnout scores for mothers, according to dimension (KES): Memory: 2.3 Physical symptoms: 1.8 Sleep disturbance / fatigue: 2.1 Emotions: 2.4</p> <p>Mean burnout scores for fathers, according to dimension (KES): Memory</p>
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					Mean burnout scores for fathers, according to dimension (KES) and condition of the child: ** Memory: 2.8 IBD Physical symptoms: 1.7 DM 1.8 IBD Sleep disturbance / fatigue: 2.3 DM Emotions: 2.4 DM		2.3 Physical symptoms: 1.4 Sleep disturbance / fatigue: 2.0 Emotions: 2.0
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* Significant $p < 0.05$

** only significant differences are presented in the table

Parental Burnout Inventory (PBI)

Maslach Burnout Inventory (MBI)

Karolinska Exhaustion Scale (KES)

Burnout Scale (BS)

Shirom – Melamed Burnout Questionnaire (SMBQ)

Stakeholders recognized that parents of children with CCN have a harder time than parents without children with CCN. Individual differences within the groups were explained by some parents having more resources to deal with challenges than others and by contextual factors. Three themes were derived from the reflections from stakeholders. The first theme encompassed the extra caregiving tasks due to children's conditions and being exhausted from having to perform physical caregiving tasks. The second theme regarded re-experiencing continued mourning with no foreseeable end (i.e. chronic sorrow, see Lindgren, Burke, Hainsworth & Eakes, 1992), as highlighted by a parent: *"What I really believe is an important theme is chronic sorrow, being continuously occupied with saying goodbye to the expectations and hope that you have for your child. Over and over again, accepting that your child cannot do 'this' and will not do 'that' or that the gap between the child's peers keeps getting bigger."* The third theme was the lack of understanding among family members, friends, healthcare professionals or other members of the social circle of parents, about what it entailed to be a parent of a child with CCN.

3.4. Prevalence

Ten articles investigated the prevalence of burnout (Table 2). Half of the studies were conducted in Sweden. To measure burnout, six studies used the SMBQ, three studies used the MBI or a derivative of it, and one study measured a broader concept 'Carer's Need Assessment for Schizophrenia' (CNA-S). CNA-S covers 18 problem areas, of which one problem area concerns burnout. The prevalence ranged from 20-77% among parents of children with CCN, and 20-34% among parents of children without CCN. However, a clear cut-off point was not always justified. The three studies using a reference sample all used the SMBQ and applied the same cut-off score.

Table 2. Prevalence of burnout among parents of children with CCN

Author (year)	Country of publication	Diagnosis/ condition of the child	Inventory used to measure burnout (reference), number of items, number of subscales, (number of items per subscale), Likert scale, mean or sum score, interpretation	Cut-off point	Sample size parents of children with CCN (n)	Prevalence of burnout among parents of children with CCN according to gender of parent/ condition of the child/ sub dimension of burnout inventory	Sample size reference parents (n)	Prevalence of burnout among reference sample
Lindström, Åman, and Norberg (2011)	Sweden	Type 1 Diabetes Mellitus	SMBQ (Melamed et al., 1999), 22 items, 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score of ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 142 mothers n = 109 fathers	37.5% all parents 44.4% mothers 28.4% fathers	-	-
Nasir (2019)	Iraq	Type 1 Diabetes Mellitus	MBI after modification (Lee, 2017), 3 subscales.	Mild mean score (1-1.66), moderate mean score (1.67-2.33), high mean score (≥ 2.34).	n = 140 mothers n = 60 fathers	23.5% mild burnout 60.0% moderate burnout 16.5% severe burnout Prevalence according to the dimensions of MBI: DP 26.0% mild 74.0% moderate PA 24.0% mild 47.0% moderate 29.0% severe EE 11.0% mild 69.0% moderate 20.0% severe	-	-

Lindström et al. (2010)	Sweden	Type 1 Diabetes Mellitus and Inflammatory Bowel Diseases	SMBQ (Melamed et al., 1999) 22 items, 4 subscales (8-4-4-6), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score of ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 163 mothers n = 125 fathers	36.0% all parents 42.9% mothers 27.0% fathers Prevalence according to diagnosis/ condition of the child: Type 1 Diabetes Mellitus: 44.4% mothers 28.1% fathers Inflammatory Bowel Disease: 33.3% mothers 17.6% fathers	n = 73 mothers n = 51 fathers	20.2% all parents 20.5% mothers 19.6% fathers
Wancata et al. (2008)	Austria	Schizophrenia	CNA-S (Wancata et al., 2006) 18 problem areas (of which one problem area concerns burnout). The severity of each problem area is rated according to a 3-point scale: no/ mild-, moderate-, or severe problem.	Based on those scoring moderate or severe on the CNA-S.	n = 101 mothers n = 101 fathers	54.8% mothers 34.6% fathers	-	-
Lindahl Norberg and Forinder (2016)	Sweden	Children who had undergone paediatric hematopoietic stem cell transplantation	SMBQ (Melamed et al., 1992; Shirom, Westman, & Carel, 1997), 22 items, 4 subscales, 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 156 mothers n = 123 fathers	34%	-	-

Riva et al. (2014)	Sweden	Children who had undergone hematopoietic stem cell transplantation	SMBQ (Melamed et al., 1992; Shirom et al., 1997), 22 items, 4 subscales, 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 146 mothers n = 114 fathers	34% all parents 39% mothers 27% fathers	-	-
Lindahl Norberg et al. (2014)	Sweden	Children who had undergone paediatric hematopoietic stem cell transplantation	SMBQ (Melamed et al., 1992; Melamed et al., 1999), 22 items, 4 subscales, 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 159 mothers n = 123 fathers	39.0% mothers 27.6% fathers	n = 237 mothers n = 166 fathers	31.6% mothers 19.9% fathers
Lindahl Norberg (2007)	Sweden	Children who had survived a malignant brain tumor	SMBQ (Grossi et al., 2003; Melamed et al., 1992), 22 items, 4 subscales (8-4-4-6), 1-7 Likert scale, mean score, higher scores indicate more burnout symptoms.	A score ≥ 3.75 indicates burnout (Grossi et al., 2003)	n = 24 mothers n = 20 fathers	54% mothers 20% fathers	n = 152 mothers n = 107 fathers	34.0% mothers 19.0% fathers
Cantwell-Bartl and Tibballs (2017)	Australia	Hypoplastic left heart syndrome	MBI (Maslach, Jackson, & Leiter, 1996).	-	n = 16 mothers n = 13 fathers	77.0% low level of burnout	-	-
Jaramillo, Moreno, and Rodríguez (2016)	Colombia	Down Syndrome	MBI (Maslach et al., 1997; Pelsma, Roland, Tollefson, & Wigington, 1989), sub dimension Emotional Exhaustion, 9 items, 0-6 Likert scale, higher score indicates more burnout.	A score > 19 on EE indicates burnout.	n = 103 mothers n = 103 fathers	52.6%	-	-

Shirom–Melamed Burnout Questionnaire (SMBQ) consisting of four subscales: emotional exhaustion/physical fatigue, listlessness, tension and cognitive difficulties. Maslach Burnout Inventory (MBI) consisting of three dimensions: Emotional Exhaustion (EE), Lack of Personal Accomplishment (LPA), Depersonalization (DP). Carers' needs assessment for schizophrenia (CNA-S) questionnaire covering 18 problem areas, of which one problem area concerns "Burn-out or illness of the carer".

Stakeholders considered the findings on prevalence as validation that parents who go through this experience do not have to feel alone in being a rare exception. Prevalence studies were seen as 'recognition' and 'acknowledgement' of the importance of this topic. Stakeholders speculated about a potential gap between research and awareness by professionals of parents' struggles. From the perspective of parents, their needs were insufficiently acknowledged and addressed. Recognizing the feelings of parents, professionals pointed out that the fragmented organization of care hampers addressing these needs.

Stakeholders indicated that the prevalence numbers would receive more value if the context of the parents of children with CCN would be described. This was reflected through the stakeholders' prominent interests in the antecedents and the consequences of burnout related to the prevalence numbers. Furthermore, stakeholders pointed towards the limited number of countries included, the heterogeneity of operationalization, and the uneven attention towards the different conditions that are associated with CCN.

3.5. Associated factors

Forty-five of the included studies investigated factors associated with burnout (Supplement 4). The majority of these studies employed a quantitative research design ($k = 35$), followed by a qualitative research design ($k = 8$), and a mixed method design ($k = 2$). We coded these factors according to the Informal Caregiving Integrative Model (ICIM) by Grain and Zech (2019) to provide an overview (Figure 3). Most quantitative studies investigated demographic factors such as parent or child characteristics (age or gender of parent and child, level of education, and family composition), or psychological factors (parent depression and anxiety). Less emphasis was placed on more distal variables such as the social context of the parent, stress and coping, and the role of healthcare professionals. The qualitative studies focused on the experiences of parenting a child with CCN.

Figure 3. Associated factors found in the literature grouped according to ICIM

Recognizing most of the extracted factors from the literature, stakeholders concluded that there was not one evident factor causing burnout. Rather, burnout stemmed from a combination and interaction of factors based on factors perceived as important to a parent. Stakeholders also emphasized that parents vary in the meaning they ascribe to what seemingly may be the same contextual factor. Unable to prioritize factors perceived as important, was considered a factor causing burnout. Stakeholders also emphasized the importance of both instrumental and emotional support provided through the formal and informal social network.

Stakeholders highlighted two important themes lacking from the literature. The first theme concerns 'incidental factors' and can be seen as 'the straw that broke the camel's back'. Parents of children with CCN are in a fragile position, where an additional incident may trigger burnout. The incidental factor does not have to concern the child with CCN and its impact is highly dependent on individual and situational factors. The second theme concerned the relationship between the parent and the child, which was impacted by several factors such as, the long-term dependence of caregiving tasks to the child, behavioural problems of the child (such as aggression), and the potential lack of reciprocity in the relationship.

4. Discussion

Considerable variation exists in how studies have conceptualized burnout among parents of children with CCN, with a majority of studies taking a domain-general approach while more recent work is focusing more on domains-specific formulations. The latter approach reveals the most pronounced contrasts between parents of children with and without CCN. Indeed, parents of children with CCN have on average more burnout symptoms compared to parents of children without CCN, and burnout is more prevalent among parents of children with CCN compared to parents of children without CCN. However, variation in conceptualization and operationalization hampers synthesis of findings on risk and resilience factors. The pattern of findings across studies as well as the conclusions from the stakeholder consultation point towards multifactorial explanatory models, in which no specific risk factors or burdens may cause burnout, but their subjective and context-specific meaning, accumulation, and interaction determine outcomes.

The synthesis and stakeholder analysis emphasizes the interaction between demands from parenting and caregiving, where parents of children with CCN may be disproportionately burdened at the expense of experiencing fulfilment in being a parent. Stakeholders mentioned exhaustion of caregiving tasks, chronic sorrow and lack of understanding and support as relevant themes. The burden of this double role is confirmed in a meta-analysis that reported small to medium effect sizes for burnout on familial caregiving towards a spouse or a child (Gérain & Zech, 2021). However, less is known about the associated factors in the context of parental caregiving burnout. A fuller understanding of the interactions between parenting and caregiving may be achieved when future studies conceptualize burnout within the specific domain of parenting and caregiving. A conceptualization highlighting this double role held relevance and face validity to the stakeholders. Although the specific context of parental caregiving burnout is different, the nature of symptoms seems to be comparable to some aspects of occupational and parental burnout (Mikolajczak et al., 2018). Furthermore, it is plausible that the consequences of

burnout affects domains beyond the domain where it originated from (Maslach & Jackson, 1981). Therefore, next to domain specific research, the broader literature on risk and resilience factors on burnout may also provide relevant insights for parents of children with CCN.

No dominant factor was identified in the literature or in stakeholders' views that may on its own explain burnout. Stakeholders emphasized that rather than on objective factors, focus should be on subjective factors that determine the burden and parents' appraisal of having the resources to cope. This is in accordance with the dynamic interplay among risk and resilience factors as outlined in the ICIM by Gerain and Zech (2019). However, this model lacks factors that are specific for parenting a child with CCN. According to the multi-dimensional theory on burnout (Maslach, 1998), these specific factors are relevant because these may determine which of the social interactions are likely to cause more burden or bring more relief. As such, future studies may also focus on gaining more understanding of the dynamic interplay between subjective experiences and objective risk factors within the context of burnout in parents of children with CCN. This may help raise awareness and attention for burnout in parents, and inform tailored interventions to parent's needs.

The strength of this study lies in its participatory approach. A recent review on the engagement of patient partners in synthesis reviews, shows that meaningful involvement enhances the relevance and quality in reviews (Boden et al., 2021). This engagement was further extended by a stakeholder consultation, which enhanced the meaning of the insights taken from the data. However, not all stakeholders were able to partake in the whole process and at times the majority of the stakeholders consisted of parents. Another limitation stems from the COVID-19 pandemic, which forced the stakeholder consultation to be conducted online. This may have led to disproportionally including stakeholders with better access to technology. However, online data collection has also logistical benefits, which may limit attrition (Sy et al., 2020). Furthermore, as our search strategy was limited to titles and abstracts, potentially relevant studies may have been missed. However, as we searched key terms relating to 'parents' and 'burnout', we feel confident that our search captured a broad scope of the literature pertaining to burnout among parents of children with CCN.

Although the exact prevalence of burnout among parents of children with CCN is unknown, parents who experience burnout within this group are far from alone. Societal awareness, especially in general and clinical practice, may be a first step towards alleviating some of the strain that parents feel (Abdoli, Vora, Smither, Roach, & Vora, 2020). Furthermore, integrating care for families of children with CCN may also alleviate some of the factors giving rise to parental burnout. For both parents and healthcare professionals it would be useful to have an actionable theoretical model explaining risk and resilience factors and consequences of burnout. A model based on data and lived experience can provide

insight in the situation of parents and points of reference for support. Such a model may help specify the benefits that may be expected from prevention and early identification, and to investigate how such benefits might be achieved. Building such a model is also urgently needed to formulate the potential benefits of clinical formulation and intervention with respect to parental burnout.

Key Messages

1. Burnout among parents of children with CCN is not a rare phenomenon, therefore awareness and particularly acknowledgement and recognition among these parents in the clinical practice is necessary.
2. Parental burnout likely stems from an individual confluence of factors that are subjectively perceived as weighing on parents, complicating the provision of support.
3. The context of parents of children with CCN is challenging due to the double role of parenting and caregiving and therefore cannot be immediately reduced to models derived from job-related burnout.
4. When interpreting research on burnout among parents of children with CCN, the variation in domains of burnout and operationalization need to be taken into account.
5. There is a need for an actionable theoretical model of the causes and consequences of burnout among parents of children with CCN.

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Conflict of interest

The authors report no conflicts of interest. The authors alone are responsible for the content and writing of the paper.

Ethics statement

Ethical approval was obtained from the Scientific and Ethical Review Board (VCWE #2020-147) of the Faculty of Behavioural and Movement Sciences, Vrije Universiteit Amsterdam.

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