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Unsung heroes, flying blind

- a metasynthesis of parents' experiences of caring for children with special health care needs at home

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Abstract

Aims and objectives: To aggregate, synthesize and interpret qualitative research studies of parents' experiences of caring for a child with special health care needs at home.

Background: Advances in the field of medical and nursing science have ensured better survival rates for children with chronic illnesses. Many of these children have significant special health care needs. Today parents assume a caregiver role, undertaking tasks previously provided by nurses in hospitals. As the complexity of care delivered by parents continues to develop, synthesized knowledge can provide an evidence base that will support and guide nurses when caring for these families.

Design: Metasynthesis.

Methods: Based upon a systematic search protocol a structured literature search, covering the years 2003-2016 was conducted in five electronic databases. Ten studies were included and appraised using the Critical Appraisal Skills Program assessment tool. A metasummary and a metasynthesis were undertaken guided by the metasynthesis methodology as described by Sandelowski and Barroso (2007).

Results: The results were interpreted and integrated under the overarching theme "unsung heroes, flying blind", supported by eight elucidating categories that illustrate aspects of the parents' life world.

Conclusions: The enormous burden of care can weaken the parents' will to carry on and result in a decreased ability to provide care. This can have an impact on the parents' health, family functioning and the sick child's potential health outcomes. Nurses are in a unique position to help these families and should be better prepared for the role.

Relevance to clinical practice: Knowledge of how parents of children with special health care needs experience their daily lives can promote trust in nurses and guide them in their efforts to support families with children living with chronic illness.

Keywords: children, special health care needs, community, parents' experience, nursing, metasynthesis

What does this paper contribute to the wider global community?

- Nurses must acknowledge the needs and expertise of parents who have children with special health care needs.
- Parents face considerable challenges. Lack of acknowledgment and support from health care professionals can exacerbate their uncertainties, worries and helplessness in their caregiver role.
- Parents' expressions of essential life phenomena such as hope, satisfaction, fatigue, shame and guilt can easily go unnoticed by health care professionals. It is important to listen attentively to each individual parent and pay particular attention to the metaphors they use.

Introduction

Advances in the field of medical and nursing science have ensured better survival rates for children with chronic illnesses (Elias & Murphy, 2012). International prevalence and incidence rates for this population group are unknown (Carnevale, Rehm, Kirk & McKeever, 2008). However, it is suggested that almost 20% of children in the United States have significant special health care needs that require care beyond the needs of healthy children (Child and Adolescent Health Measurement Initiative, 2012). A minority of these children have multiple, complex needs, requiring medical technology and continuous help from health care professionals (Elias et al., 2012). Children with special health care needs are defined as “those who have or are at increased risk for a chronic physical, developmental, behavioral, or emotional condition and who also require health and related services of a type or amount beyond that required by children generally” (McPearson et al., 1998). In order to delimit the metasynthesis, this research study focuses on children with physical conditions who require care beyond the needs of healthy children. The formidable growth of managed care, and the move towards more primary health care, mean that these children receive treatment and care in their homes (Rehm, 2013). This implies that parents assume a caregiver role, undertaking caregiving tasks previously provided by nurses in hospitals (Caicedo, 2014).

Background

Previous metasyntheses and systematic reviews have investigated parents' experiences of caring for children with chronic illness (Coffey, 2006; Kepreotes, Keatinge & Stone, 2010; McCann, Bull, & Winzenberg, 2012). These studies show that parents of children with special health care needs experience both positive and negative emotions related to a considerable burden of care involving rigid routines and little flexibility. Parents, especially mothers, report sleep problems, fatigue and anxiety related to constant monitoring and worrying about their sick child (Carnevale et al., 2008; McCann et al., 2012). Social support and well-functioning care services are of great value in coping with caring responsibilities (Rehm & Bradley, 2005). Smith, Cheater and Becker (2015) carried out a rapid structured review of the literature on parents' experiences of caring for children with special health care needs and recommended a metasynthesis of parents' experiences. A metasynthesis of the multitude of studies on caring for the very sick child can enhance knowledge in the field (Bondas & Hall, 2007). This review can help to fill the knowledge gap.

Nurses have developed a variety of practice models to guide generalist and advanced practice with families (Wright & Leahey, 2013). Foster, Whitehead, Maybee and Cullens (2013) recommend research into parents' views on what can shape positive and negative health care experiences. Research has demonstrated that nurses do not always conduct care in a respectful manner, acknowledging parents' knowledge and expertise (Shields, Pratt, & Hunter, 2006). The needs of the parents, family members and sick child can vary, but parents play a key role in coordinating care and initiating care needs (Macias, Clifford, Saylor & Kreh, 2001). Recognizing the pivotal role of parents as advocates for the entire family is important. When parents struggle, the whole family struggles (Smith et al., 2015). Previous research highlights the importance of ensuring that parents' needs are met (Årestedt, Benzein & Persson, 2015). Knowledge about how these parents manage their daily lives can support nurses and guide them in their efforts to alleviate suffering and promote trust for the whole family. This knowledge can also guide nurse educators in their teaching role, and ensure that the needs of parents and families are taken into consideration, when developing curricular plans.

The aim of this metasynthesis is to aggregate, synthesize and interpret existing qualitative research studies of parents' experiences of caring for a child with special health care needs at home, in order to improve clinical nursing practice. The research question addressed is: "How do parents experience caring for a child with special health care needs at home?"

Methods

A metasynthesis is a third person interpretation of primary qualitative studies. “Metasyntheses are integrations that are more than the sum of parts in that they offer novel interpretations of findings that are the result of interpretive transformations far removed from these findings as given in research reports” (Sandelowski & Barroso, 2007, p.18). The aim is “to generate a form of knowledge that is enriched by the different disciplinary angles of vision, methodologies, samples and interpretive lenses each original investigator brought to the challenge” (Thorne, 2017, p.3). Findings from metasyntheses have the potential to promise a generalizable theory that can be transferred to clinical nursing and policy makers in the field (Finfgeld-Connet, 2009).

Research design

The study followed Sandelowski & Barroso’s’ (2007) methodological procedures, which combine technical standards with a creative approach, and where the findings are synthesized into a metasummary and a metasynthesis. A figure and tables were used to provide an audit trail, and enhance transparency. To reach consensus and promote trustworthiness throughout the process, the authors worked separately in appraising, classifying and synthesizing the findings, before working together to reach consensus after each phase.

Data collection

Based on a systematic search protocol, a medical librarian conducted a broad and structured literature search in five databases in June 2016: PubMed, Cinahl, Scopus, Cochrane and Embase. The **Population, Interest, Context (PICO)** tool (Joanna Briggs Institute, 2014) helped formulate the research question and facilitated the search process. The PICO elements are illustrated in Table 1. The inclusion criteria followed logically from the PICO concept grid. Table 2 provides an overview of the inclusion and exclusion criteria.

Table 1 Clarification and structuring of research question using PICO

Table 2 Inclusion and exclusion criteria

Keywords and word stems from the different PICO elements constituted the search strategy. Combinations and variations of subject terms, thesaurus and free text searching were modified to fit the different database search systems. Between November 2016 and January

2017 database searches were supplemented with additional searches: a “backward search” in reference lists, and a “forward search” in “cited by” columns in databases, including Google Scholar.

After duplicates were removed, the searches produced 618 electronic publications. Titles and abstracts were read. Papers not meeting the inclusion criteria were filtered out and the remaining abstracts (n=125) were reviewed, 44 articles were selected for inclusion. Of 44 full text articles, 34 were excluded, as they did not meet the criteria for inclusion. Ten articles from 2003- 2016 describing factors affecting parents caring for children with special health care needs in primary care were included for quality appraisal. The flow chart (Figure 1) illustrates the search strategy.

Figure 1 Flow chart

Appraising and classifying findings

Critical appraisal of the studies included in the analysis is fundamental to prevent the inclusion of inadequately reported studies. To ensure the quality of each primary article, the Critical Appraisal Skills Program (CASP, 2017) tool was used. This instrument consists of 10 questions that help evaluate the congruity between aims, methods, design, data collection, analysis, findings and discussion in the included studies. The authors appraised the studies independently. Disagreements were resolved through discussion. No study congruent with the aim of the metasynthesis was excluded due to poor quality.

The CASP tool does not provide a scoring system. It does however provide the authors with a checklist for appraising the evidence. The appraisal of the evidence is always based on thorough and judicious discussions concerning the quality of the evidence retrieved. Judgments of the studies depends on what the primary authors’ report (Sandelowski & Barroso, 2007). The form and content of the included studies were appraised. Based on the CASP appraisal all 10 articles were included as illustrated in Table 3.

Table 3 CASP, checklist for appraising qualitative studies

Most of the data were interpretive and descriptive, with findings presented in themes, concepts and topics, close to the participants’ own words. Such interpretive explanations address the fundamental nature of experiences, as this method focuses on variations in

sampling, data and the target phenomena (Sandelowski & Barroso, 2007). Some of the included articles were highly descriptive in nature, and provided more insight than others. The characteristics of each study are shown in Table 4.

Table 4 Characteristics of the included studies

Analysis

Following Sandelowski and Barroso (2007), the findings were synthesized using two approaches: metasummary and metasynthesis. A metasummary is an aggregation of the findings with a calculation of their frequency and intensity. Metasummaries are useful when searching for patterns in the data and serve as an iterative process that guides consensus in the analysis (Ludvigsen et al., 2016). This method provided an empirical basis as it demonstrated how much emphasis should be accorded each category in the synthesized findings. Concepts borrowed from the primary studies (in vivo concepts) were used to categorize the findings. Effect sizes were calculated (Table 5). Effect sizes are quantitative transformations of qualitative data in order to show how the findings are weighted and to verify the presence of patterns or themes (Sandelowski & Barroso, 2007).

A metasynthesis of abstracted findings was carried out using reciprocal translation analysis (Sandelowski & Barroso, 2007). In vivo concepts, such as original metaphors, concepts, and phrases that conveyed essential and existential meanings, were used to systematize, and group the findings. The process is illustrated in column 1, of Table 6. Quotations from parents (first-order constructions) and/or primary researchers' interpretations of parents' experiences (second-order constructions) were extracted. These were further abstracted into eight categories, as illustrated in column 2, of Table 6.

The studies were sufficiently analogous to enable a reciprocal translation analysis of the findings from one study to another. Qualitative research rarely results in complete congruence of meanings, and allowing for differences provides an understanding of other perspectives and interpretations. An iterative process of interpretation, was carried out, to review important connections and patterns among the first- and second-order constructions, and create third-order interpretations and is shown in column 3, of Table 6. In this approach, metaphors, themes, concepts and contexts are revisited and compared across studies. The process is always influenced by the metasynthesis researchers' personal reflections.

Findings

The findings are reported as a metasummary and a metasynthesis and supported by tables. Table 5 illustrates the intrastudy effect sizes and interstudy frequency sizes of each category. Table 6 provides an overview of the in vivo concepts and the second- and third-order analyses

Table 5 Metasummary of intrastudy effect sizes and calculated interstudy frequency sizes of every category

Table 6 In vivo concepts, second-order and third-order analysis

Metasummary

The 10 included studies, described in Table 4, were conducted in urban and rural areas in Australia (1), the UK (2), Taiwan (3, 6), Norway (4, 9), Sweden (7, 10) and the USA (5, 8). The data collection methods used in the studies were individual- (1- 10) and focus group interviews (1, 4), based on strategic or purposeful sampling. The methods of analysis used were qualitative and descriptive. The authors in nine of the ten studies were nurses. The word *parents* is used for the participants who had the primary caregiving responsibility for the children. In all, 120 mothers, including 12 foster mothers and one adoptive mother, and 52 fathers comprised the sample. The parents' education, occupations and socioeconomic background varied. Some of the parents were divorced and others were single mothers. The ages of the children involved ranged from eight months to 17 years and they had a broad range of illnesses that demanded special health care. The metasummary, illustrated in Table 5, describes the frequency and effect sizes for each finding. Almost all the studies are represented in all categories. This strengthens the validity of the metasynthesis.

Second-order analysis

Eight categories emerged from the synthesis (second-order analysis). Each category is illuminated by rich quotations from parents caring for children with special health care needs in the 10 studies. The categories, as listed in column 1 of Table 6, are described in more detail in the following paragraphs. The numbers in parentheses refer to the study from which the in vivo concept was extracted.

Flying blind

All 10 studies reported that parents daily face unpredictable situations, especially related to the continuous changes in the child's disease, needs and behavior, described in one study as an “emotional rollercoaster” (Hayles, Harvey, Plummer, & Jones, (2015, p. 1142). The metaphor “flying blind” is quoted in (Hayles et al., 2015, p. 1143) and alludes to the unpredictable life situation parents experience.

Referring to the unpredictable changes in the child's health, many parents felt uncertain about how to interpret symptoms of possible illness progression and how to act accordingly. Parents in nine of the ten studies felt unprepared, because no one had told them what to expect. “We just winged it year, by year” (Hayles et al., 2015, p. 1143). “Nobody’s actually turned round and said, right these are the symptoms and these are what you should look for.” (Hinton & Kirk, 2016, p.5).

For parents, it was frustrating not to have sufficient experience to distinguish clearly between symptoms of illness and the child's own distinctive personality.

“The child, he lives in danger all the time. You really need to get to know the child.

They give you cues, but if you don’t know the language, didn’t learn the language, you can’t respond in the right way, and that can be detrimental” (Lauver, 2008, p.85).

Acute life-threatening episodes were described in several studies as especially unpredictable. An example of this is asthma attacks at night, when the parents were terrified because they did not know what to do if the child was unable to breathe, and might die in their arms. “... I felt so anxious about him” (Trollvik & Severinson, 2004, p.95). Parents of children with epilepsy described similar feelings. Epilepsy seizures made them highly stressed because they did not know what to do to remedy the situation (Mu, 2006, p. 547). Many had learned the effective strategy of taking one day at a time and solving problems as they arose, since it was impossible to plan their days.

Always at the back of my mind

Constant worry was discussed in all primary articles. Parents distinguish between everyday worries and worries about the future. They worry about the progression of the disease and their inability to act: “It’s always at the back of my mind”. “You worry about everything, that’s the burden you carry around with you” (Sallfors & Hallberg, 2003, p. 198). Parents also had difficulty letting go of the sick child, due to their reluctance to hand over the responsibility for medical care to others.

The parents had almost no leisure time, as many monitored the child continuously: “It's just like you are tense and you cannot relax at all. Just like waiting for a war, very nervous” (Mu, 2006, p.546). Concerns tended to escalate at night, and disrupted sleep was an explicit finding in several articles. One mother said:

I have this image of a ship going through a very narrow strait with very high sharp rocks on either side and you can hit something if you go ... at night-time you never really know what it is going to hit (Sullivan- Bolyai, Deatick, Grupposo, Tamborlane & Grey, 2003, p. 25).

For some parents, the concerns decreased as they began to feel more secure and experienced as carers, but for others concerns persisted: “I was never at ease with my son's illness, and I am still not. I think it's frightening” (Trollvik & Severinson, 2004, p. 95).

A repeated finding in most studies is concerns about the children's futures: “Will my child be able to walk? Will my child ever go to school? What will it be to grow up? Will he be able to have a girl friend?” (Sallfors & Hallberg, 2003, p.199). Parents also worried about who would take care of their child when they grow old or die (Kvarme, et al., 2016, p. 366).

A further concern was the child's future ability to cope with studies, work and moving away from home (Hinton & Kirk, 2016, p. 6). The concerns seemed to increase when parents met other children with similar diagnoses or progressing disease. As one parent said:

I used to get MS charity magazine for him, but I've stopped that as well now because I felt as though everyone was bed ridden and I thought I can't cope with this” (Hinton & Kirk, 2016, p. 9).

Over time, the parents realized that the best strategy was to focus on what actually brought joy to their days, rather than pessimistic concerns about a future they could not predict anyway. (Hinton & Kirk, 2016, p. 9).

Managing the frontline - a constant battle

All articles confirmed that parents carry an enormous responsibility for giving children with special health care needs the best possible care. This responsibility includes understanding the child's health condition, coordination and transportation to and from health appointments, giving information to health professionals, school care and administering necessary medications and medical procedures. One foster mother said:

I just wanted to love her, help her progress, and get her obstacles, but it turned into a challenge, not one we were necessarily up to. Who'd have thought we'd be doing what we are doing for her today? Takin' care of the Broviac (catheter), TPN (total parenteral nutrition), G-tube, teachin' home nurse how we do things (Lauver, 2008, p. 90).

The parents described their responsibility as overwhelming and time-consuming, and felt unprepared for the amount of responsibility they were obliged to take on. Tight structures and careful planning were important to balance every day. No matter how well days were organized, parents found that unexpected situations “always” arose. Patience and flexibility to address sudden incidents were an absolute necessity:

Changing diapers is quicker with a normal child, but in my case it takes about an hour just to put him down, arranging him, and take off his diaper (Kvarme et al., 2016, p. 366).

To be best prepared to meet the child's needs, parents even took on the responsibility of navigating the health care system, special-interest organizations and the Internet for information.. For some, navigating gave a sense of control over the situation:

I decided that before I stuffed that new medicine into my child, I'd check in the physician's desk manual. When we got to the hospital, I make them check...everything (...): You have to be calling them all constantly, and asking questions. Otherwise things can go wrong (Sallfors & Hallberg, 2003, p. 200).

Navigating the system was a source of frustration and powerlessness, “...we don't know who to approach (...). And how long to wait before we should be getting some direction on what we should be doing...it's really difficult...” (Hayles et al., 2015, p.1145).

They described a continuous struggle:

It's been a constant fight and battle. I'm still fighting to get a standing frame at school...then it was a constant battle or fighting for therapy...it doesn't matter what we do, we have to fight and challenge everyone and everything (Hayles et al., 2015 p.1146).

Parents (5, 7- 10) found that the challenges related to the child's special needs strengthened family ties and gave new perspectives on what was meaningful in their lives.

Somebody to share my responsibilities with

Support from family, friends, people in similar situations, and district nurses was conveyed in all articles as crucial for motivation, relaxation and relief in parents' everyday lives. Support was given in many ways, ranging from emotional dialogues to practical assistance such as housekeeping or babysitting. The support prevented parents from feeling completely alone:

...I feel warm.... somebody to share my responsibilities. They show concern about me and didn't blame me" (Mu, 2006, p.549).

Older siblings were considered a support as they knew the sick child well and could report symptom changes. Parents also had good support from each other, even though one article reported broken marriages directly related to the complexity of their situation (Kvarme et al., 2016).

Communication and support from health professionals was evident in the findings, but only a few studies discussed experiences directly related to nurses. In many articles, parents reported needing help from clinicians. They could not manage without this support. Recognition and acknowledgment from health professionals gave them encouragement and confidence in their role as caregivers for their sick children and was often more important than information on symptoms and practical assistance: "...they do not give us anything more than peace of mind, so to speak" (Wennick, Lundgvist, & Hallström, 2009, p. 227).

However, many parents had negative experiences of meetings with health care professionals. They described being ignored and mistrusted, which triggered stress and a sense of wanting to give up. The quality of the relationship with health care professionals was considered essential. Distrust of professionals in critical situations was also frightening, and led to parents losing all respect for health care providers:

We were advised to call the hospital when our child started wheezing. We know more about our child than a physician who meets him once a year (Trollvik & Severinson, 2004, p.96).

Parents were expected to learn complex procedures and skills quickly in order to master the child's needs. Healthcare professionals lacked understanding of what was required to deal with the challenges: "Yeah, they give you a bag of supplies and say: Okay, take her home, you'll be fine, right" (Sullivan-Bolyai, et al., 2003, p. 25). Also:

"I think sometimes the health care professionals needs to be around the family a bit more...the parents need to be cared for a bit more...and something to embrace with other siblings, because we get tired, we get stressed..." (Hayles et al., 2015, p.1147).

Another mother said: “.... We don’t want to feel like a number or just another case” (Hayles et al., 2015, p. 1145).

It was all my fault

Parents in all of the articles, described guilt regarding the sick child, the siblings and the family’s reputation. Essentially, mothers take the blame for the child's restrictions in everyday life. They feel responsible for having brought the child into the world and guilt because the child has inherited the disease and the suffering from them: “I was shocked when my child took ill. I was so sad because I myself suffer from allergies. I found it so depressing that she developed the same disease” (Trollvik & Severinsson, 2004, p. 95). Another mother said: “She will turn fifteen next year...sometimes she would scold me that I caused her CP, that it was all my responsibility” (Huang, Kellett, & St. John, 2011, p. 192).

Parents also blame themselves for having living conditions that could potentially exacerbate the child's disease: “If I had cleaned the house properly she would not have become ill” (Trollvik & Severinsson, 2004, p. 96). The mothers felt guilty because they had less time to spend time with siblings: “I felt sorry for my eldest son, because I ignored his feelings and needs since I needed to give all my attention to caring for the child with CP” (Huang, et al., 2011, p.192). Another mother described doing everything to make the sick child feel confident, at the expense of siblings who also needed to be seen:

...my son questioned why he should always let her do what she wanted to do. I said no one would give her first priority, if you didn’t” (Huang, et al., 2011, p.192).

Making the most of their life

The phenomenon of hope is expressed in different ways in nine out of ten articles, and is seen in close relation with the parents’ concerns. The parents described different pathways to hold on to hope and maintain an optimistic outlook: “I believe things will work out. I hope our child will outgrow it. You need to be positive” (Sallfors & Hallberg, 2003, p. 198). Some parents hoped that second opinions from doctors about the child's condition would provide a better prognosis, while others wished for a breakthrough in research and new treatment options. They contacted organizations and specialists in the hope of encouraging responses: “I suppose we are all looking for the same thing, we’re looking for someone to say: Oh we got a cure” (Hinton & Kirk, 2016, p. 7).

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It gave parents hope to experience that others with the same disease mastered the situation (Sallfors & Hallberg, 2003, p.200). Another strategy for preserving an optimistic outlook was to avoid contact with people, even close family, who sabotaged their aspirations: “So I basically said, you know, I am not going to tell you. So we stopped telling him” (Hinton & Kirk, 2016, p.8).

Consistently the parents aspired for the child to be able to live a normal life in terms of their own possibilities. The parents therefore hoped that the child would become sufficiently independent to master everyday life and that the illness would not worsen. Many parents wished others could see the child and not just the disease. As one parent said referring to her daughter:

I am a little girl who’s got an unusual situation because I have all these people involved...but I am a little girl. I am not a patient all the time and having to do all this is just...a very small part of who I am (Hayles et al., 2015, p. 1148).

Parents prayed that the child would have the same opportunities as other children and tried to treat the sick child as they did the healthy siblings: “We kept things as normal as possible” (Sallfors & Hallberg, 2003, p. 200). This was not always easy, as the disease, demanded a rigid structure governing everyday life and restrictions on activities.

Suffering in silence

It is evident from the articles (2- 4, 6- 10) that parents experienced feelings of shame at having a child with special health care needs, and tried to hide the child from family members, but also that family members tried to hide the child from others to avoid harm to the family’s reputation:

“My mother-in-law was hiding my child with CP in order to avoid our relatives” (Huang et al., 2011, p.192).

Parents were afraid that others had negative pre-conceptions of the social connotations of the disease: “In the past it was a negative stigma about epilepsy, - Afraid children will laugh at her at school” (Mu, 2006, p. 546). The child's special needs were consequently a taboo that parents avoided talking about. Many parents chose to not inform others, not even their immediate family or the child's teacher about the disease, because they feared that the stereotypical attitudes would harm the child. Some mentioned stigmatizing events from school:

Her class mates now think she is different and she is feeling isolated. One day, she told me tearfully that she had no friends so she didn't want to go to school (Huang et al., 2011, p. 193).

It could be difficult to convince others that the child was sick and needed special consideration. Parents were therefore reluctant to be open about the situation: "Sis, you told us that she is ill, but she doesn't look ill to us" (Hinton & Kirk, 2016, p. 6).

In some articles, parents longed for their children to be regarded as just as smart as healthy children, despite their limitations: "He is so smart, very intelligent and very sweet ... He is very positive, always smiling and laughs a lot" (Kvarme et al., 2016, p. 364).

On duty all the time

Most parents admitted to having health challenges directly caused by the enormous care burden the child's special needs entailed. The high care burden over time led to many physical ailments: "I used to carry her around she is heavier now so I have pain in my hands and back" (Kvarme et al., 2016, p. 366). Mothers as primary caregivers were especially vulnerable. One mother said that she had to "wear a mask" and pretend that life was going smoothly to prevent losing all control of the situation.

Sleep disorder was reported as a significant problem in several studies (4, 6, 8, 9, 10). A mother's plight was described: "She (mother) is crying all day long, she cannot sleep at all. She is almost burned out" (Mu, 2006, p. 547). Many articles reported declining emotional health such as depression, loneliness and the feeling of isolation (4, 6, 7, 8, 9). The mothers' health, especially among single mothers, was most affected. Most fathers denied any health problems. In all articles, the mothers had the primary caring responsibilities for the child's special needs, while the fathers worked outside the home. Parents felt that home nurses and other forms of relief were essential for physical and mental health.

Third-order analysis

The secondary analysis provided a "reconceptualization" of the primary findings at a higher level of interpretation, which facilitated a third-order analysis as described by Sandelowski and Barroso (2007). The third-order analysis revealed that parents' struggle and their courage to carry on and face challenges is supported by very few life-promoting phenomena. Life-constraining phenomena dominated their experiences.

Metasynthesis: unsung heroes, flying blind

A metasynthesis should be presented in a way that captivates the audience and faithfully represents the lives of the participants (Sandelowski & Barroso, 2007). Taking into consideration the complexity of the phenomenon studied, this metasynthesis is achieved by integrating and interpreting phrases and metaphors from the primary studies. A more comprehensive understanding of parents' experiences when caring for a child with special health care needs is created.

The metaphorical phrase: "unsung heroes, flying blind" characterizes and brings together the various findings related to the parents' experiences. An underlying tone in the data is that the parents use war metaphors as a rhetorical strategy to bring forth their experiences. War terms such as "struggle", "fight", "battle", "defend" and "frontline" are consistently, used to communicate a situation that is difficult to put into words. This metaphorical language is further reinforced in descriptions such as "flying blind", "we winged it", "sleeping with our eyes open", "vigilant caregiving", "waiting for a war", "fighting spirit" and "it ricochets back at you".

Lakoff and Johnson (1980, p. 455) explain that "the essence of metaphor is understanding and experiencing one kind of thing or experience in terms of another". It is interesting that parents use war images to portray and structure their experiences. Although there is no visible physical battle, a metaphorical and existential one appear to exist. We use the meta-metaphor "unsung hero, flying blind"- to reflect the invisibility and unpredictability of the parents' struggle. Wars can be won or lost by following certain strategies but the parents do not have the weapons to provide the best defence. Usually, war metaphors are used against the illness itself (Sontag, 1978), but in these data the parents also struggle with health care professionals.

Discussion

To our knowledge, this is the first metasynthesis offering a new and comprehensive understanding of what it means to be a parent of a child with special health care needs at home. Admittedly, Coffey (2006) published a metasynthesis on parenting a child with chronic illness, but the study is almost 12 years old, and was in the authors' own words "a compilation of experiences of parents who care for children requiring medications, treatments, and technology support every day of their lives" (Coffey, 2006, p. 58). This metasynthesis is both a compilation and reciprocal translation of findings in the studies examined, which through the methodology produces a new interpretive perspective of the

parents' experiences less well explored in previous research. Kepreotes et al., (2010) synthesized experiences of parenting children with chronic health conditions up to the year 2007, and identified that caring for children with complex chronic health conditions required further investigation.

The findings are similar to previous reviews indicating that long-term illnesses in children influence parents' lives socially, psychologically and physically, as well as challenging their resources for coping and reconciliation (Coffey, 2006; Cousino & Hazen, 2013; Fischer, 2001; Kepreotes et al., 2010; McCann et al., 2012; Smith et al., 2015). However, the approach in this study, with its focus on existential life phenomena, is new.

Discussion of findings

The overall findings show that it is not just the disease itself that creates worries for the family. How the parents understand and make sense of the illness is as much of a challenge. This provides new insights into how existential life phenomena, such as *hope*, *trust* and *the courage to carry on*, are constrained by feelings of *guilt*, *shame* and *worry*. The synthesis demonstrates a deeper understanding of what it means to balance an existence in which life phenomena constantly and unpredictably fluctuate between being life-promoting and life-constraining. Life phenomena come to the fore when serious illness strikes a family (Delmar, 2013). Nurses who work with these families must be attentive to parental needs to prevent essential life phenomena, as expressed by the patient and family, from being overlooked and ignored. Martinsen (2006) writes about the importance of promoting life when suffering is intense.

To have somebody to share responsibilities with is pivotal in enabling parents and preventing them from feeling completely alone. Support from others is not always available (Rehm & Bradley, 2005). The complexity of the child's special health care needs made it difficult for parents to accept support, resulting in social isolation (Kirk & Glendinning, 2005).

Hope as a life-promoting phenomenon is present in most of the studies in the analysis. Although the understanding of hope is subjective, a common feature is that hope is expressed as a specific and concrete possibility for improvements in the child's health. Hope has implications for how parents experience their present situation. Similar to these findings, Duggleby et al. (2010) revealed that hope among family caregivers of persons with chronic diseases is a dynamic experience of possibilities within a framework of uncertainty. As long as there is hope of overcoming adversity, parents continue to request second opinions from

physicians and navigate the search for additional information or new research results. The parents found meaning in small improvements and took one day at a time. These findings are consistent with Dufault and Martocchio's (1985) description of the contextual dimension of hope. Continual adjustment in order to live a normal life is a hope pathway for many parents. The findings show that parents deliberately avoid situations that might be potentially embarrassing and cause shame. Shame is a dominant phenomenon in the primary articles from Taiwan. Parents strive for an everyday life with strict procedures aiming at minimizing problems (Rehm & Bradley, 2005).

Life-constraining factors refer to phenomena that limit the parents' ability to make their own choices, and that can increase their suffering and challenge well-being in their daily lives. There is evidence confirming that the daily lives of parents with chronically sick children are constantly constrained by ongoing worries, overwhelming parenting responsibilities, and an enormous care burden (Caicedo, 2014; McCann et al., 2012).

In this synthesis the overarching metaphor "flying blind" alludes to the unpredictability of the parents situation in knowing how to deal with the sick child's evolving care needs. Parents experience acute life-threatening situations and are often "flying blind", as the only thing they can rely on is their own intuition. The parents in Coffey's (2006) metasynthesis use the metaphor: "tricks of the trade" to describe the evolving needs of the sick children on a daily basis.

Consistent with previous research (Fischer, 2001; Nuutila & Salanterä, 2006), it is clear that parents of chronically ill children have an overwhelming desire for updated knowledge and information from healthcare professionals. In addition, earlier studies (Coffey, 2006, Keprekotes et al., 2010) show that parents have a strong desire for predictability in terms of being more involved in the child's care and treatment than these findings have revealed. The findings indicate, however, that mutual trust and respect from healthcare professionals strengthens the parents' self-esteem and gives them the courage to carry on. This corresponds with Wright and Bell's (2013) findings that families experiencing caring relationships with healthcare professionals are less likely to feel isolated, vulnerable or uncertain in their caregiving role. Nurses have the potential to recognize and make visible the multiple ways in which life phenomena are expressed in meetings with parents of children with special health care needs. Nurses have an ethical and a moral responsibility to promote life and soften suffering. This support can be essential for the parents' will to carry on, in spite of the adversity they face. A situational ethical approach that is sensitive to the uniqueness of each situation is therefore a prerequisite for good practice.

Accepted Article

It emerged from the findings that health-care professionals do not always seem to understand the parents' situation and are not always receptive to their experiences. Parents want to be more than just a "number in the line". Martinsen (2006) writes about the importance of nurses having a personal professional nursing approach. The nurse must listen attentively to the parents and ask relevant questions that can reveal their values, beliefs and experiences. This can pave the way for dialogue. A true dialogue between the parents and nurse may be the most important way of promoting mutual trust and respect.

Being the mother of a child with special health care needs is associated with a higher risk of fatigue and illness (Caicedo 2014; Mc Cann, Bull & Winzenberg, 2015). The fact that mothers often take the main responsibility for care can explain the gender differences in health problems. A mother with poor health can cause the family further strains (Hauge et al, 2013; Leiter, Krauss, Anderson & Wells, 2004). Nurses should map resources and possibilities for respite, as well as considering interventions that promote hope and meaning.

Evidence from previous research reveals that mothers, in particular, experience strong feelings of guilt. They blame themselves for being responsible for the sick child's reduced well-being (Jackson & Mannix, 2004). The findings suggest that nurses need to recognize these feelings of guilt, and support the mothers in their mothering role.

The metasynthesis shows that feelings of *guilt*, *shame* and *worry* can be life-constraining factors for the parents. Findings from prior studies have shown that perceived stigma can affect well-being and reduce the willingness to socialize with others (Goffman, 1963; Green, 2003).

The parents' use of war-metaphors indicate a form of warfare against both the child's illness and an intangible and at times inadequate system of care. These findings support the study's overriding metaphor, *unsung heroes, flying blind*. Lakoff and Johnson (1980) maintain that metaphors have the strength to formulate reality. The implied message, being that parents, mothers in particular, express that their life situation as comparable to a state of emergency. They are on constant standby in order to maintain a balance in life and to protect themselves and their families from a relentless foe. If they are not adequately prepared, they risk defeat and capitulation. Metaphors can have a "feedback" effect (Lakoff & Johnson, 1980). In this case, the parents' use of war metaphors can influence interactions with nurses. Meeting a parent on the warpath can result in the nurse becoming defensive, which can again reinforce the parents' feeling of fighting constant battles in which they view the health professional as an opponent and not an ally.

Acknowledging the parents' use of war metaphors can be key to understanding their experiences and provide a starting point for building relationships based on respect and trust. Martinsen (2006) says that trust is a surrender of oneself, whereby the other receives the trust shown and thereby acts. Listening attentively to the parents' illness-experiences is the most beneficial nursing intervention (Årestedt et al., 2015). This evidence should be of the utmost importance to nurses in primary care, a field in which requirements for advanced family care are greater than ever.

Discussion of method

Bringing together and summarizing the findings in a metasummary before performing a metasynthesis strengthens the credibility of the research (Sandelowski & Barroso, 2007).

Despite having carried out a comprehensive search strategy, with the support of a librarian, potential studies may have been overlooked as no search strategy guarantees full coverage. The effect sizes would have been different had the searches been limited to databases. Additional searches in reference lists and citation tracking led to data saturation and increased the validity of the synthesis. This search strategy revealed data sufficient for a metasynthesis of the in-depth experiences of 172 parents of children with special health care needs. A possible limitation is that 'grey' literature was not included.

Preserving the raw data through direct quotations in the findings promotes validity. In studies with few quotes, there was difficulty in distinguishing first- and second-order interpretations. Furthermore, it was difficult to determine the extent to which the primary researchers' interpretations were influenced by their background and epistemological stance. Another possible limitation is the predominance of mothers over fathers in the interviews. Transferability is both strengthened and limited by the various cultures and contexts of the original studies. Cultural differences can affect how parents feel supported. Different health care systems and variations in services and help can also influence the parents' experiences. Nevertheless, studies from different parts of the world ensure the diversity of findings and increase understanding of the complexity of the research topic. Studies using different designs and approaches were included, based on the argument that the findings are more important than the methods of data collection and analysis (Bondas & Hall, 2007). A well-documented audit trail was maintained throughout the process and creative interpretations, which surpass rigid methodological forms of assessment, were incorporated, thus strengthening the credibility of the synthesis (Sandelowski & Barroso, 2007).

The first author has worked as a nurse in a hospital setting with very ill patients and their families for many years. The second author has long clinical experience in primary care, as a public health nurse, working with children and families and subsequently as a qualitative researcher in the field. The authors are aware of the danger of over-interpretation of findings when researching a familiar area. The authors have sought transparency throughout the study and have provided thick descriptions of findings from the primary studies.

Research implications

Findings in this metasynthesis show a striking predominance of life-constraining factors in parents' experiences, calling for examination of nursing interventions to support the parents' caregiving role. Further research should focus on the underlying meanings of parents' war metaphor experiences, their situated possibilities, their worries and what really matters in their lives. The importance of dialogue with health professionals is evident in the findings. The reason for the reported lack of dialogue between clinicians and parents is unclear and could benefit from further research. Although paediatric home care is favored in caring for children with special health care needs, there is currently little research on the impact of this on parents, siblings and the sick children. Such studies should include parents' socio-economic background, family structure, culture, and age of the children. The phenomena of *shame* is more dominant in the Asian articles. Our findings have shown that shame can cause isolation and deter families from seeking support; this should be an area for further research. The long-term health consequences of parenting a child with special health needs have not been adequately documented, despite widespread recognition of the huge emotional distress and long-term strain involved (Caicedo, 2014; McCann et al., 2012).

The research strategies for exploring parents' experiences of caring for children with special health care needs at home are dominated by semi-structured interviews to elicit participant responses. Source triangulation, focus group interviews, field observations, action research or web-based research might add nuances to research perspectives and create awareness of professional and parental roles and responsibilities. These research approaches can support the need for continued education and clinical supervision of nurses in complex care practices. A meta-analysis of the extent, distribution and magnitude of certain quantifiable findings, such as the effect and type of health problems these parents experience, could expand this research. Researchers should also consider whether studies should involve the family as a group or as individual members. Studies of parents' experiences of chronically sick children seem to favor the recruitment of mothers. There is limited research

on fathers' and siblings' experiences. Sick children's perspectives could also be an area for further research.

We recommend user involvement in future metasyntheses research. Collaborating with representatives from community nursing, nursing education and families can be vital for formulating relevant research questions and ensuring continued reflection throughout the research process.

Conclusion

This metasynthesis brings together an interpretation and integration of findings from 10 qualitative studies that increase our understanding of parents' experiences of caring for a sick child at home. A significant feature of caring for these children, independent of diagnosis and across cultures, is the considerable challenge faced by parents, as well as the lack of acknowledgment and support from health care professionals, which exacerbates their uncertainties, worries and sense of helplessness in their caregiver role.

The ship metaphor was mentioned by a mother in one study. If a boat trip is a metaphor for how parents' experience their life situation, a reflection on who should comprise the crew is vital. A morally responsible way of helping people in trouble at sea is to navigate the ship towards a safe harbour. Even small adjustments in the course can make significant difference on whether the ship remains afloat. Nurses are in a unique position to be part of the ships' crew, offering safe navigation routes and making sure there is adequate safety equipment on board for the whole family.

Relevance to clinical practice

Parents' expressions of essential life phenomena such as hope, satisfaction, fatigue, shame and guilt can easily go unnoticed by health care professionals. This study is relevant to clinical practice and can serve as a reminder that listening to each parent and the metaphors they use is important. Listening with an open and attentive attitude is required to preserve their integrity, boost their confidence and provide opportunities for reflection. This is especially important as parents may be at risk of depression, exhaustion, anxiousness, sleep deprivation and loss of hope. As the complexity of parents' tasks in the home increases, we argue for knowledge-based practice in which nurse practitioners and researchers share knowledge so that parents of children with special health care needs can benefit from continual knowledge development. In a health policy context, the actors involved should stand together and raise developments with health politicians, also where necessary initiating

a change of practice. The chance of success increases further if the nursing care of children and families has a more dominant place in nursing education, ensuring that nurses are adequately prepared to meet the needs of parents with a sick child at home.

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Table 1 Clarification and structuring of research question using PICo

Research question:	<i>“How do parents experience caring for a child with special health care needs at home”</i>
Population	Parents who have children with special health care needs
Phenomena of Interest	Gain knowledge about the everyday experiences of parents who have children with special health care needs
Context	Parents caring for children with special health care needs, at home

Table 2 Inclusion and exclusion criteria

Inclusion criteria	Exclusion criteria
<ul style="list-style-type: none">• Published primary articles in English• Research from all countries for the period 2003 to 2016• Qualitative methodology• Research on parents' experiences from their everyday lives with children with special health care needs, that have implications for nursing• The first person voice of the parent(s) had to be adequately represented	<ul style="list-style-type: none">• Studies of children with mental disorders/diagnoses, due to the heterogeneity of the patients sampled• Research focusing on children with palliative diagnoses, or other diagnoses requiring advanced care, as we anticipated that the daily life of these parents was different• Studies using quantitative methodology• Systematic reviews and metasyntheses were excluded

Table 3 CASP, checklist for appraising qualitative studies

First author, year	Was there a clear statement of the aims of the research?	Is a qualitative methodology appropriate?	Was the research design appropriate to address the aims of the research	Was the recruitment strategy appropriate to the aims of the study?	Was the data collected in a way that address	Has the relationship b/t researcher and participants been adequately considered	Was the data analysis sufficiently rigorous?	Have ethical consideration been taken into consideration?	Is there a clear statement of findings?	How valuable is the research?
Hayles (2015)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Hinton (2016)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Huang (2011)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Kvarme (2016)	Y	Y	Y	Y	Y	Y	Y	CT	Y	Y
Lauver (2008)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Mu (2006)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sallfors (2003)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Sullivan-Bolyai (2003)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Trollvik (2004)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y
Wennick (2009)	Y	Y	Y	Y	Y	Y	Y	Y	Y	Y

Y= yes - N=no - CT= can't tell

From: Critical Appraisal Skills Programme (2017).

Table 4 Characteristics of the included studies

Number	First Author, year	Country of origin	Aim	Research design	Method	Setting & sample	Data analyses	Major Findings
1	Hayles, 2015	Australia	To explore parents' experience of health care for their children with cerebral palsy living in a regional area of Australia	Grounded Theory	Focus group and individual in-depth interviews	Setting: Rural hospital, by telephone Sampling: 13 parents of children with CP aged less than 17. (11 mothers, 2 fathers, including 1 foster mother and 1 adoptive mother)	Constant comparative analysis	*Parents experience health care as a "cyclical process" of: -making the most of their body and their life. -learning as you go-navigating the systems-meeting needs through partnership-being empowered or disempowered-finding a balance
2	Hinton, 2016	United Kingdom	To explore the experiences of parents of children with multiple sclerosis	Grounded Theory	Semi-structured interviews	Strategically sampling Setting: Participants homes Sampling: 31 parents (20 mothers, 11 fathers)	Constant comparative method	*Dominated of feelings of uncertainty:-diagnostic uncertainty-daily uncertainty-interaction uncertainty-future uncertainty
3	Huang, 2011	Taiwan	To describe a range of challenging caregivers experiences for Taiwanese mothers providing care for their children with cerebral palsy (CP)	Qualitative hermeneutic phenomenological	In-depth audio taped interviews with each mother	Purposeful sampling Setting: not mentioned Sampling: Mothers who were primary caregivers for children with CP. Criteria based recruitment. n=15, 27-42 year	Hermeneutic analysis, guided by Heidegger and Gadamer	*Being concerned:- Experiencing burden as a sole primary caregiver-managing balancing demands-being marginalized by others-Encountering limited or no professional support
4	Kvarme, 2016	Norway	To provide knowledge about how immigrant parents of children with complex health needs manage their family lives and how this affects their own health and quality of life	Qualitative study, exploratory design	Individually and focus group interviews	Purposive sampling Setting: Not mentioned Sample: 27 parents, 18 mothers and 9 fathers from Pakistan (n=13), Poland (n=9) and Vietnam(n=5)	Kvale and Brinkmanns phenomenological hermeneutic guidelines	*Immigrants parents of children with complex health needs experience their own health and quality and life challenges. -Pain, sleep disturbance - mental health problems - lack of respite care, social network, support*Mothers were the primary care giver*Struggle to access resources for their child
5	Lauver, 2008	USA	To explore the experience of parenting foster children with illness and complex medical needs	Qualitative, Phenomenological	Unstructured interview	Purposive sampling Setting: Participants home Sampling: 13 fosterparents. 7 mothers and 3 couples.All fosterchildren had multiple health problems	Van Manens methodology	*Parenting a chronically ill foster child with complex medical needs is a multifaceted experience having implications for multiple disciplines*Being committed to the child*Coming to know the needs of a medically complex foster child*identifying effective and ineffective interventions
6	Mu, 2008	Taiwan	To investigate the essence of the family health-illness transition experience from the parental perspective when a child is affected with epilepsy	Phenomenological	In-depth interviews	Purposive sampling Setting: not mentioned Sampling: Inclusion criteria. 8 couples, 2 mothers.The children age range from 3-6.8 year	Colaizzi's approach	*Being emotionally traumatizes and physically exhausted*Vigilant parenting*Reflected family resilience as the most important resource *Facing the social challenge*Pain associated with the stigma of epilepsy

7	Salfors, 2003	Sweden	To explore parents' experience of living with a child with Juvenile Chronic Arthritis	Grounded theory	Open interviews	Strategically sampling Setting: in the informants home, at the clinics and at a conference hotel Sampling: 22 parents (6 fathers) of children aged 7-17 years with Juvenile Arthritis	Constant comparative method for Grounded theory	*Deeper understanding of experienced parental stress in daily living with a chronically ill child-Parental vigilance-Emotional challenges-Continual adjustment*Reflects traditional gender based parenting roles, complementary and necessary for the family *
8	Sullivan-Bolay, 2003	USA	To provide a detailed description of day-to-day management experiences of mothers raising young children under 4 years of age with type 1 diabetes	Qualitative, Naturalistic inquiry	Open interviews	Setting: in the mothers home Sampling: 28 English-speaking mothers of children under 4 years of age, diagnosed with diabetes for at least 3 months	The NUD*IST software program (1997)	The central theme was constant vigilance. The mothers described having to use hyper vigilant caregiving behaviour to accomplish the day-to-day management. Mothers described three aspects of constant vigilance: - day-to-day concerns- day-to-day management- supportive resources
9	Trollvik, 2004	Norway	To describe nine parents' everyday experiences of living with a child suffering from asthma	Qualitative, descriptive	Semistructured interviews	Setting: Seven parents were interviewed at their homes, two at the hospital Sampling: nine parents of five children with moderate to severe asthma in the 2-6 years age group	Phenomenological content analysis	Findings divided into 4 themes: *Feelings of uncertainty, helplessness and guilt*Need for support and help from healthcare professionals*Adaption to everyday life
10	Wennick, 2009	Sweden	To illuminate the everyday experience of family members 3 years after a child had been diagnosed as having Type 1 diabetes	Qualitative	Individual interviews	Setting: chosen by the participants, mostly their own homes Sampling: 35 familymembers with a child having Type 1 diabetes (5 affected boys, 6 affected girls, 3 male siblings, 1 female sibling, 9 fathers, 11 mothers) Inclusion criteria	Latent content analysis	*Illness being deeply rooted into their everyday life, but influenced different family members in different ways- Ambivalent parenthood- Stressful daily planning- Knowledge and tried experience skills- Capricious blood sugar level- Physical activity as pleasure and annoyance- Fear of losing control- A private and public concern

Table 5 Metasummary of intrastudy effect sizes and calculated interstudy frequency sizes of every category

v	Categories (abstracted findings)	Flying blind	Always at the back of my mind	Managing the frontline – a constant battle	Somebody to share my responsibilities with	It was all my fault	Making the most of their life	Suffering in silence	On duty all the time	Intrastudy intensity effect size
	Author, year									
1	Hayles, 2015	X	X	X	X	X	X			75% (6 of 8)
2	Hinton, 2016	X	X	X	X	X	X	X	X	100% (8 of 8)
3	Huang, 2011	X	X	X	X	X	X	X	X	100% (8 of 8)
4	Kvarme, 2016	X	X	X	X	X		X	X	87.5% (7 of 8)
5	Lauver, 2008	X	X	X	X	X	X			75% (6 of 8)
6	Mu, 2006	X	X	X	X	X	X	X		87.5% (7 of 8)
7	Sallfors, 2003	X	X	X	X	X	X	X		87.5% (7 of 8)
8	Sullivan-Boyai, 2003	X	X	X	X	X	X	X	X	100% (8 of 8)
9	Trollvik, 2004	X	X	X	X	X	X	X	X	100% (8 of 8)
10	Wennick, 2009	X	X	X	X	X	X	X		87.5% (7 of 8)
	Interstudy frequency effect sizes	100% (10 of 10)	100% (10 of 10)	100% (10 of 10)	100%(1 0 of 10)	100% (10 of 10)	90% (9 of 10)	80% (8 of 10)	70% (7 of 10)	

Table 6 In vivo concepts, second-order and third-order analysis

In vivo concepts	Second-order interpretations	Third-order interpretations
Flying blind (1)	The child's needs are increasingly changing Unpredictable everyday life Learning by doing Little guidance to manage the situation	Existential life-conducting phenomena -Promoting self-expression -Nurses are in a unique position to alleviate suffering and promote healing in these families, and should be better prepared for this role. Existential life-constraining phenomena -Inhibiting self-expression -Less courage to persevere in parents may decrease their ability to care for their child with special health care needs
Always at the back of my mind (7)	Constant worries Continuous monitoring Future concerns	
Managing the frontline (7)	Overwhelming responsibility Time-consuming tasks Took responsibility to retrieve information Mothers had primary responsibility at home Stronger family ties Deeper understanding of others' suffering	
Somebody to share my responsibilities (6)	Essential coping factor Support from family, friends, health professionals, spouse and siblings Practical assistance Emotional support Disrespectful, distrustful, not very appreciative meetings with health professionals Good meetings valued, supportive Important that the whole family was considered	
Making the most of their life (1)	Hope for a stable future Hopes of normalization Hopes for a breakthrough in research	
Suffering in silence (6)	Disease is a taboo they don't talk about Hiding the child Stigmatization by others	
On duty all the time (4)	Health challenges related to stress and overwhelming tasks all the time	
It was all my responsibility (4)	Try to pretend that everything is okay Taking on guilt for the child's illness and sufferings Difficulty in reconciliation Guilt for siblings	

(The numbers in parentheses in column one refer to the study from which the in vivo concept was extracted)

Figure 1 Flow chart

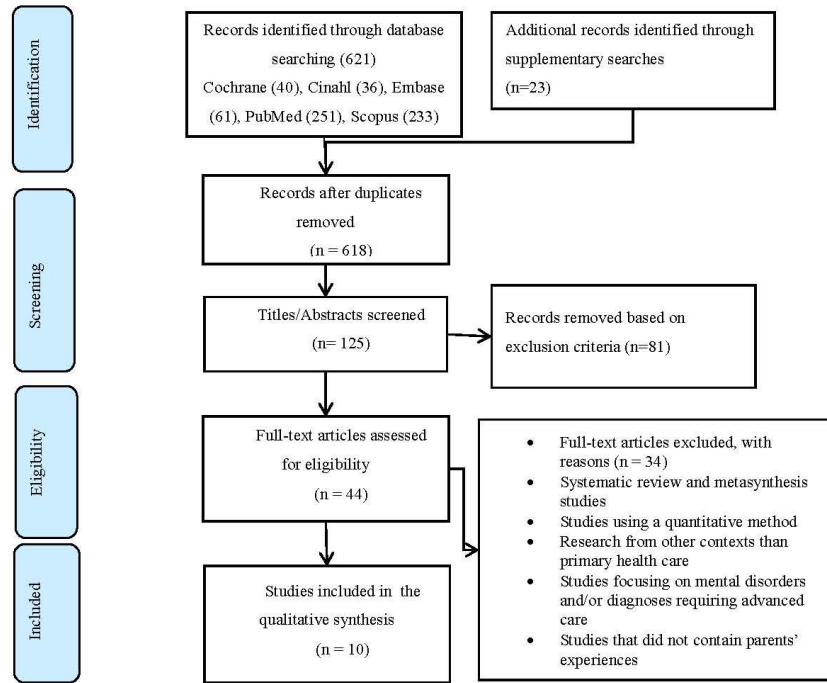


Figure 1: Flow chart of systematic literature search From: Moher D, Liberati A, Tetzlaff J, Altman DG, The PRISMA Group (2009). Preferred Reporting Items for Systematic Reviews and MetaAnalyses: The PRISMA Statement. PLoS Med 6(7): e1000097. doi:10.1371/journal.pmed1000097