A meta-ethnography of parents' experiences of their children's life-limiting conditions

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Abstract: As many children's life-limiting illnesses (LLCs) are now often viewed as curable, there is an inevitable tension between providing good treatment and addressing patients and their families' needs. For healthcare providers to provide optimal care, they must understand parents' experiences of illness. Therefore, this article provides a meta-ethnography of parents' experiences of their children's LLCs by examining the findings of existing IPA studies. Seventeen studies were included, which allowed the development of a conceptual model. Two multifaceted concepts emerged from the data, namely *living in a bounded and polarised space* and *living in a collapsed time*, and these are discussed with reference to their sub-concepts. Recommendations for future research and practice are provided.

Keywords: IPA, meta-ethnography, life-limiting, parent experience, children, healthcare

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1. Introduction

The Royal College of Paediatrics and Child Health (RCPCH), in collaboration with the UK-based Association for Children's Palliative Care (ACT) defines the concept of life-limiting conditions (LLCs) as those for which there is no reasonable hope of cure and impose limits on a person's quality and/or quantity of life (RCPCH/ACT 2009). Life-threatening conditions

are those for which treatment may be feasible, but can fail. Hain et al. (2013) developed a directory of life-limiting conditions that comprises a list of nearly four hundred ICD10 codes associated with diseases that can limit life in children. A condition is defined as life-limiting if its course can potentially be described by at least one of the archetypes set out in the ACT/RCPCH (2009) guidelines: (a) conditions for which treatment may be feasible but can fail or when palliative care may be necessary due to unsuccessful treatment or during an acute crisis, irrespective of the duration of the threat to life (e.g. cancer, cardiac anomalies, irreversible organ failures of heart, liver, kidney from anorexia, coeliac disease-related morbidities as oropharyngeal and oesophageal cancers); (b) conditions where premature death is inevitable but there may be long periods of intensive treatment aimed at prolonging life and allowing participation in normal activities (e.g. Duchenne Muscular Dystrophy, cystic fibrosis); (c) progressive conditions without curative treatment options, where care is solely palliative and commonly extends over many years (e.g. metabolic or neurodegenerative conditions) and (d) irreversible but non-progressive conditions causing likelihood of severe disability and premature death through complications (e.g. severe cerebral palsy, multiple disabilities following brain or spinal cord injury, complex health care needs and a high risk of an unpredictable life-threatening event or episode). These conditions have varying morbidity, however they share the commonality of probable reduced lifespan and possible or certain early death. While this classification serves the purpose of planning and needs assessment, not all children in these four groups need active palliative care throughout the trajectory of their medical condition. For example, some children in the second group, may have long periods of relatively good health and whilst being seriously disabled may not need active palliative care. Others may need active palliative care from an early stage (RCPCH/ACT 2009).

LLCs render the child or young person increasingly dependent on parents and carers even if palliative care is not provided. As for many life-limiting illnesses the treatment is now often viewed as feasible, there is an inevitable tension between providing good treatment and addressing patients and their families' needs. The difficulty of the treatment process lived simultaneously with the hope of relief from the disease often leads to conflicting psychological states among patients and their caregivers.

One of the aims of interpretative phenomenological analysis (IPA) in healthcare is to understand and make sense of the experience of illness, and the complex processes involved. IPA has been used extensively within health psychology. Recently there has been a growing corpus of IPA research examining parents' experiences of their children's LLCs. However, this research proliferation can make it difficult to use this knowledge in practice. Since the physical impact of the diagnosis is seen as an immediate target for intervention, diagnostic and treatment protocols frequently omit to address how children's diagnoses might impact on their parents' experiences as caregivers. Additionally, it is difficult for busy healthcare practitioners to review the extensive literature (Burgess et al. 2005). Recent data suggest an increasing incidence of paediatric LLCs (Collins et al. 2016). There is a growing need to support caregivers who tend to their children's healthcare needs for extended periods of time. Despite bearing a heavy responsibility for the child's personal and nursing care (RCPCH/ACT 2009), family members may cope with changes in life associated with the diagnosis (Cederborg et al. 2011; Eatough et al. 2013). There is a need of further education of healthcare professionals and family practitioners with regard to parents' experiences of caring for a child with an LLC so that effective psychological interventions and support channels to empower them and reduce the care giving burden can be developed (Brewer et al. 2007; Eatough et al. 2013).

The aims of this meta-ethnography were: (i) to provide an in-depth assembly of the current state of knowledge around parents' experiences of their children's LLC; (ii) to understand the impact of healthcare services on parents' experiences, and (iii) to contribute to the development of methods for IPA research synthesis. By pulling together and interpreting IPA findings from across different settings, this study aimed to move beyond the description (Mays et al. 2005) of parents' experiences and healthcare services and generate a conceptual model (Tong et al. 2012) explaining what it is like to have a child diagnosed with an LLC. There has been no previous such attempt. The only identified meta-ethnography in this area of research was published by Heinze et al. (2012) and examined the end-of-life decision making of parents of children with cancer.

2. The method

A meta-ethnography approach was used to understand parents' particular experiences of their children's LLCs. Meta-ethnography is an interpretive form of knowledge synthesis, proposed by Noblit and Hare (1988) that aims to develop new conceptual or 'metaphorical' understandings. Although it has been argued that meta-ethnography is more suitable to synthesise a small number of studies (Campbell et al. 2003; Hannes et al. 2012), the majority of published syntheses using meta-ethnographic methods have included a number of studies ranging from 3 to 44 (Dixon-Woods et al. 2007; Campbell et al. 2011; Hannes et al. 2012). The method has been successfully used to synthesise qualitative studies focusing on managing anti-depressants (Malpass et al. 2009), experiences of a traumatic birth (Elmir et al. 2010), informal care for stroke survivors (Greenwood et al. 2010), breast cancer (Adams et al. 2011; Banning 2011), dementia (Seitz et al. 2012) and parents' end-of-life decision making (Heinze et al. 2012). This meta-ethnography was structured according to the Enhancing Transparency in Reporting the synthesis of Qualitative research (ENTREQ) (Tong

et al. 2012) and the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) (Noyes et al. 2015) recommendations.

2.2. Search strategy

Four electronic databases were searched: PubMed, PsycINFO, CINAHL and Science Direct. Studies were included up until September 2017. Search terms available from the InterTASC Information Specialists' Sub-Group (ISSG) Search Filter Resource (see www.york.ac.uk/inst/crd/intertasc/; accessed July 2017) were used to develop the search syntax. Most of the databases included the following key terms: (docsubtype(FLA) or docsubtype(SSU) or docsubtype(SCO)) and "Interpretative phenomenological analysis" OR "Hermeneutic" OR "Heidegger" OR "Husserl" OR "Colaizzi" OR "Strauss" OR "Van Kaam" OR "Van Manen" OR "Gadamer" OR "Ricoeur" OR "Spiegelberg" OR "Merleau" AND "qualitative research" OR "interview" OR "semi-structured" OR "in-depth" OR "face to face" OR "unstructured" OR "discussion" OR "qualitative study" OR "focus group" OR "group discussion" OR "informal" OR "fieldwork" AND "purposive sampling" AND "life experience" OR "lived" AND "life-threatening condition" OR "limiting illness" OR "diagnosis" OR "palliative care" OR "end of life" AND "parents" OR "children" AND "English language". The search was augmented with hand searches of articles cited in reference lists and from relevant review papers.

2.2. Study selection

Studies were included if they were (i) IPA studies exploring parents' experiences of their children's LLCs and (ii) full text papers published in peer-reviewed journals. Studies were excluded if they (i) did not include parents' experiences; (ii) used other methods than IPA; (iii) children's illnesses were not life-limiting; (iv) explored parents' experiences of children's death and grieving; (v) were not published in English language. In selecting the

relevant papers, the archetypes set out in the ACT/RCPCH (2009) and the directory of LLCs developed by Hain et al. (2013) were used. Studies that explored parents' experiences before and after the child's death were included if findings had been reported separately.

2.3. Data screening and extraction

Search results were collated in an Excel database and duplicates deleted, followed by initial screening of titles. A priori inclusion/exclusion criteria were applied at this stage. Abstracts of any titles retained were then screened. A second researcher (MM) screened the abstracts and was involved in the quality appraisal of full-text papers. Any discrepancies were resolved by consensus.

Data extracted from the articles included study details, such as: author, year of publication and country, participants' characteristics (number, gender and age), response rate, source of participants, method and time of data collection, and children's characteristics (diagnosis, age, number and gender). Full replication of two key types of data as specified by Noblit and Hare (1988) was used: (i) the literal participants' experiences expressed in original quotations in the papers (first- order data), and (ii) authors' interpretations and conceptualizations (second-order data). In studies where data collection took place after the child's death (e.g. Reilly et al. 2008; Popejoy 2015), only data relating to parents' experiences of illness while the child was alive were extracted.

2.4. Quality appraisal

The Critical Appraisal Skills Programme (CASP) checklist (CASP 2010) was used to identify and appraise methodological quality of the included studies. The quality appraisal process was concerned with study rigor, credibility and relevance of the research question to the meta-ethnography topic (Hannes et al. 2011). A second researcher (MM) was involved in the quality appraisal. An agreed numerical score was assigned to each CASP question to indicate

whether it had (1) not been addressed, (2) been addressed partially or (3) been extensively addressed. Each paper was given a score ranging from 10 to 30. However, studies were not excluded from the review on the basis of the critical appraisal as a particular CASP cut-off point has not been established for ratings of quality of qualitative studies (Campbell et al. 2003; Atkins et al. 2008; Pound et al. 2005). Nine studies were of high quality (Bezance et al. 2014; Brewer et al. 2007; Eatough et al. 2013; Glasscoe et al. 2008; Glasscoe et al. 2011; Reilly et al. 2008; Thomson et al. 2014; Ware et al. 2007; Wright 2017). The remaining, while judged lacking in some quality, were still considered to constitute valuable research and contribute to the existing knowledge.

2.5. Analysis

The Noblit and Hare's (1988) seven stages of the meta- ethnography approach were used: (1) getting started; (2) deciding what is relevant; (3) reading the studies; (4) determining how studies are related to each other; (5) translating studies into each other; (6) synthesising translations and (7) expressing the synthesis. A table using first- and second- order constructs as rows and papers as columns was created to facilitate reciprocal translation. This helped identifying whether constructs corresponded ("reciprocal synthesis"), contradicted each other ("refutational synthesis") or expressed different aspects of the topic under study (a "line of argument synthesis") (Saini and Shlonsky 2012). For example, if one study reported that parents were struggling with little or no support from the healthcare team (Brewer 2007), and another that they were gaining support (Wright 2017), these were considered refutational and translated into a new construct incorporating elements of both (e.g. 'hospital as a polarised space'). However, if a third study (Hannan et al. 2005; Ware et al. 2007) suggested parents found it hard to receive support initially but shifted over time to seeking, and finally receiving support from the healthcare team, all three interpretations (Brewer 2007: little or no support; Wright 2017: receiving support; Hannan et al. 2005; Ware et al. 2007: shift from not

receiving to seeking support) were treated as reciprocal and translated into a construct encompassing all three. Sub-constructs were also developed; for example, 'getting external legitimacy' was subdivided into 'trust and shared decision making' with the healthcare team (Glasscoe 2011; Popejoy 2015), 'resolving or avoiding conflict with care providers' (Thomson 2014) and 'furthering education and learning the medical language' (Schweitzer et al. 2012; Reilly et al. 2008). These helped reconfirm the construct names in the final list and explore the lines of argument. Preliminary overarching third- order constructs from the final list of first- and second- order constructs evolved. All third- order constructs were checked back against (i) the first- and second- order constructs and (ii) the original articles to ensure accuracy and sufficiency. Table 2 describes the constructs supporting the model.

3. Results

Titles of 910 papers were screened, out of which one was a duplicate. In total, 909 titles and abstracts were screened for relevance. Of these, 829 did not meet the inclusion criteria. Therefore, 80 full manuscripts were obtained. Of these, 49 were non-IPA studies (e.g. Baird et al. 2016; Gómez-Ramírez et al. 2016; Hocking et al. 2014), three studies were systematic reviews (e.g. Xafis et al. 2015) and other three included parents of children with non LLCs, such as chronic pain (Maciver et al. 2010) and Autism Spectrum Disorder (Martins et al. 2013; Burrell et al. 2017). Two studies reported data from other family members (Cowan 2014; Sand et al. 2010), one explored the experience of death and dying and another one included an irrelevant topic. Yet another manuscript was not a journal paper. Finally, 17 papers met the inclusion criteria. The process of identification and selection of papers and PRISMA flow chart of article inclusion is outlined in Figure 1.

[figure 1 here]

3.1. Characteristics of included studies

12 studies were conducted in the UK, one in Sweden and another one in different countries of Europe; two in Canada and one in Australia. 235 parents were interviewed and data were collected through semi-structured interviews in all studies. Both parents attended the interviews in 10 studies. Six studies reported findings from mothers (Glasscoe et al. 2008; Reilly et al. 2008; Glasscoe et al. 2011; Whitehurst et al. 2011; Bezance et al. 2014; Popejoy 2015) and one from fathers (Ware et al. 2007). Children were diagnosed with different LLCs, such as: *acute liver failure* (Wright 2017), *fetal alcohol spectrum disorders* (Whitehurst et al. 2011; Coons et al. 2016a; Coons et al. 2016b), *anorexia nervosa* (Bezance et al. 2014; Thomson et al. 2014), *cancer* (Hannan et al. 2005; Schweitzer et al. 2012), *stroke* (Jones et al. 2012), *cystic fibrosis* (Glasscoe et al. 2008; Glasscoe et al. 2011), *Juvenile Huntington's disease* (Brewer et al. 2007; Eatough et al. 2013), *coeliac disease* (Cederborg et al. 2011), and others generally named *LLCs* (Ware et al. 2007; Popejoy, 2015). The study published by Reilly et al. (2008) focused on the experiences of mothers who lost a child with *an intellectual disability*. Diagnoses and causes of death included: cerebral palsy, profound and multiple intellectual disabilities, Down syndrome, global developmental delay, leukaemia, liver failure, degenerative conditions, septicaemia and epilepsy. For the purpose of this study, only the themes exploring parents' experiences when the child had been alive were used. The extracted data from each of the primary studies and second-order concepts (superordinate themes only) are presented in Table1.

3.2. A conceptual model of parents' experiences

Two multifaceted concepts emerged from the literature: (1) living in a bounded and polarised space and (2) living in a collapsed time, as shown in Figure 2. The studies all related to one another through contribution to the following key concepts.

Key concept 1: Living in a bounded and polarised space

This key concept refers to parents' caring experiences, and the interaction with healthcare professionals. Life was perceived as a paradoxical and ambivalent continuum where hospital and home were two opposite poles. Within this key concept, six concepts were identified: struggling to define boundaries of family intimacy, difficulty to get external legitimacy, lack of trust in services and disappointment, little or no direct professional support, strategies to continue life, and personal growth.

[table 1 here]

In three studies (Hannan et al. 2005; Brewer et al. 2007; Jones et al. 2012) parents described the experience of feeling less independent, due to theirs and their children polarised needs for privacy. In hospital family routine was disrupted and caring for the child, even at home, was a challenge. Visiting staff were perceived as part of the family or as intrusive (Brewer et al. 2007). Families managed their tensions by negotiating independence; while mothers experienced a strain between caring and an overwhelming urge to overprotect, fathers often wished the ill child was actively involved in the care process (Jones et al. 2012).

Six studies described parents' difficulty to get external legitimacy, as their role in the care process was questioned (Brewer et al. 2007) or their concerns were not taken seriously (Cederborg et al. 2011; Bezance et al. 2014). Parents disapproved of how their situation was contained by primary care professionals (Thomson et al. 2014) and how authorship of the child's plan for future was assumed (Glasscoe et al. 2011). However, medical team taking away end-of-life care decisions from parents released some burden from themselves (Popejoy 2015). Being part of the treatment process was essential: 'You can't ignore the parent and the parent's feelings. They have to realize how powerless you are. That actually you don't necessarily want that power to be taken, to be further eroded' (Brewer et al. 2007).

Some parents' expectations regarding care have not been fully met and six studies reported themes relating to the concept of lack of trust in services and disappointment (Ware et al. 2007; Thomson et al. 2014; Reilly et al. 2008; Hannan et al. 2005; Eatough et al. 2013; Coons et al. 2016b), while one study described parents' need of trust in the care team (Glasscoe et al. 2011). Perceived inadequacies of professionals, diagnosis and follow-up after diagnosis, poor communication across services (Ware et al. 2007; Reilly et al. 2008), parents' avoidance of conflict and a fear of betrayal by approaching services (Thomson et al. 2014) added to their stress: 'No one was meeting me in that conversation (…) And because that, because I was finding difficulties in having that conversation with professionals I was feeling I couldn't really have it with my friends or family' (Reilly et al. 2008). Parents felt that information was being withheld from them (Hannan et al. 2005). However, the relationship with the medical staff was crucial (Glasscoe et al. 2011). Although some families were very appreciative of physicians' support and service provided (Coons et al. 2016a), there was an expressed concern regarding professionals' lack of knowledge and understanding of the diagnosis (Eatough et al. 2013; Coons et al. 2016a). The need of further education was highlighted: 'Doctors and nurses…all need to be educated. Even if they think they don't need to be educated, they need to be re-educated' (Coons et al. 2016b).

Five studies reported themes relating to delayed advice (Brewer et al. 2007), struggle to access the right help (Bezance et al. 2014), the need of night-time respite and concerns around out-of-hours support (Hannan et al. 2005), lack of provision from professionals and services that understood the diagnosis (Whitehurst et al. 2011), bureaucracy, rare opportunities to discuss the diagnosis and its implications after the initial shock and insufficient support or follow-up following diagnosis (Ware et al. 2007). Knowing that a member of the healthcare team was there for them was reassuring: 'I felt like that in the darkness somebody was holding my hand' (Brewer et al. 2007). Fathers felt that

appointments were held at inflexible times and involved long periods of unnecessary waiting. They were not given equal opportunities to meet the healthcare team (Ware et al. 2007). Parents' expectations regarding how their children would die were not realized and symptoms were not properly controlled (Hannan et al. 2005).

[figure 2 here]

Eleven studies explained parents' strategies to continue life: getting external legitimacy (Glasscoe et al. 2011; Popejoy 2015; Thomson et al. 2014; Schweitzer et al. 2012; Reilly et al. 2008), gaining support (Wright 2017; Ware et al. 2007; Reilly et al. 2008; Hannan et al. 2005; Eatough et al. 2013; Schweitzer et al. 2012), advocating for children and lobbying for improved services (Ware et al. 2007; Glasscoe et al. 2008; Coons et al. 2016a) and being altruistic (Schweitzer et al. 2012; Ware et al. 2007). Some parents felt responsible for the way their children were in the world and lobbied for improved services, advocated for their children, particularly at school, and even tried to educate their children's teachers (Schweitzer et al. 2012; Coons et al. 2016a; Ware et al. 2007). Part of being able to manage their child's illness involved learning and understanding the medical terminology (Schweitzer et al. 2012). Some parents navigated on the internet to find solutions for certain situations (Brewer et al. 2007) or locate available supports (Coons et al. 2016a). Others sought to get ownership of the care plan by increasing child's autonomy (Glasscoe et al. 2008; 2011) or not allowing the school to make decisions without consulting them (Coons et al. 2016a).

Six studies outlined themes that pertained to parents' personal growth: what and how to value in life (Ware et al. 2007; Popejoy 2015) and becoming more mature, caring and positive (Reilly et al. 2008; Thomson et al. 2014; Coons et al. 2016a). Parents re-evaluated their priorities and reconsidered what was vital to them individually and as a family. There was a new sense of what happiness meant (Schweitzer et al. 2012).

Key concept 2: Living in a collapsed time

This key concept gives an account of how parents experienced loss in their inside and outside worlds. Within this key concept seven concepts were identified: alienated self (Eatough et al. 2008; Glasscoe et al. 2008; Ware et al. 2007), dilemma over competing role demands (Glasscoe et al. 2011), forgotten or diminished sense of own identity (Bezance et al. 2014; Wright 2017; Ware et al. 2007; Whitehurst et al. 2011), no sense of normality and intimacy (Bezance et al. 2014; Ware et al. 2007; Brewer et al. 2007), feeling emotionally and physically overwhelmed (Bezance et al. 2014; Wright 2017; Whitehurst et al. 2011; Ware et al. 2007; Thomson et al. 2014; Glasscoe et al. 2008; Eatough et al. 2013; Popejoy 2015; Jones et al. 2012; Schweitzer et al. 2012), no contemplation on future (Eatough et al. 2013), the paradox and ambivalence of living (Bezance et al. 2014; Ware et al. 2007; Thomson et al. 2014; Eatough et al. 2013; Glasscoe et al. 2008; 2011; Schweitzer et al. 2012; Cederborg et al. 2011; Popejoy 2015; Jones et al. 2012; Reilly et al. 2008).

Parents in three studies (Eatough et al. 2013; Glasscoe et al. 2008; Ware et al. 2007) described a feeling of self-alienation, and loss was experienced at two stages: at child's birth and diagnosis, and most relevant for this paper, the terminal stage of illness (Reilly et al. 2008). Parents suppressed their emotions or adopted a stoical attitude while dealing with loss (Eatough et al. 2013). Fathers felt the pressure to maintain a public face of being strong and masking their true feelings from others and from themselves, although privately they might have been in turmoil: 'And it is very tricky and sitting in a room with dads, all who want to appear macho' (Ware et al. 2007).

Some parents experienced the dilemma of not being able to provide treatment to their child although they were trained professionals: 'Even though I'm a nurse, I don't feel comfortable giving him his IVs' (Glasscoe et al. 2011). Diluting boundaries with their child was a difficult

task, this giving them a sense of diminished own identity (Bezance et al. 2014). Many elements of the adult world were out of parents' control (Wright 2017). This made them feel fearful and distressed (Ware et al. 2007).

Life was perceived as a battle, this leading to internal conflict. Parents found themselves in positions that were incongruous with their instinctive parenting behaviour or made them feel guilty for losing hope (Whitehurst et al. 2011; Popejoy 2015). The illness was perceived as disempowering and dehumanizing and dominated parents' thinking throughout the day. Some parents felt like a slave by sacrificing their own lives to meet their children's needs. They found it difficult to define their identity or own sense of normality and intimacy, as their lives were suddenly and irrevocably altered (Ware et al. 2007). Others felt unbounded and could not enjoy their social life together (Bezance et al. 2014) or could not keep their boundaries with the child: 'But she was becoming like a mollusc to me. It was becoming difficult to define who was her and who was me' (Bezance et al. 2014).

The emotional and physical impact of caring was overwhelming: 'I just stopped sleeping [cries]. I was a complete wreck' (Bezance et al. 2014). Some parents' worlds were 'turned upside down' before coming to terms with the diagnosis (Schweitzer et al. 2012). Others felt hopeless and powerless: 'I have lost my hope [crying]. This disease deprived me of joy of life; there is nothing that could make me really happy' (Eatough et al. 2013). There was an expressed need from parents to be cared for: 'they were our carers as well as X's carers' (Brewer et al. 2007).

Life was predominantly lived in the present, with no future plans, which very often gave families a feeling of being placed outside the social world (Eatough et al. 2013). In addition, there was a feeling of lived paradox and ambivalence. Sometimes mothers felt responsible for the way their children were in the world while other times they put it all down to fate or

chance (Glasscoe et al. 2008). Further ambiguity was experienced when parents felt guilty or responsible to make their child better themselves (Thomson et al. 2014). Although the child was still alive, chronic grief was stemming from their sense of children's loss: 'It's like you're grieving for your own child because a certain amount of your child has actually died. You're seeing it every day, you're grieving for it every day' (Jones et al. 2012). The ambivalence of living was augmented when parents were fearing or preparing for death although their child seemed well, outwardly appeared to be no different to others or did not need immediate treatment (Ware et al. 2007). Preparing for the child's death on more than one occasion was traumatic and stressful, and made future acceptance of the child dying difficult (Reilly et al. 2008).

These inner conflicts led to a pervasive sense of loneliness and isolation from support: 'I just felt that…it was just me and her. And my family were just going away from it because they didn't know how to deal with it' (Bezance et al. 2014). Isolation was experienced both within the family and from the outside world (Eatough et al. 2013), mainly when people seemed to protect themselves from the illness (Bezance et al. 2014). Family and friends were also a reason of alienation as they were perceived as failing to understand the situation and being less available or supportive than expected. Some parents felt betrayed, mainly when they perceived stigma as their friends were not comfortable to talk about their child's health-related difficulties (Thomson et al. 2014). Parents' lives and daily routine were organised around the child's care needs (Schweitzer et al. 2012), thus became painstaking and repetitive: 'I cancelled my life for her' (Eatough et al. 2013). In this battle, parents tried to make sense of their experiences, but they were unable to do so (Ware et al. 2007).

4. Limitations and strengths

4.1. Strengths and limitations of the literature

The identified IPA studies allowed for an in-depth examination of parents' experiences of their children's LLCs. This is a useful groundwork for future research and intervention. However, there was a bias toward mothers' opinions as only one study included responses solely from fathers (Ware et al. 2007). Even in studies with mixed samples, the percentage of mothers was considerably higher. Another study (Coons et al. 2016) reported on different family members, all referred to as parents. There was a lack of cultural diversity among study participants, as 14 out of 17 studies were published in Europe (12 in the UK). No study has explored the experiences of parents from minority ethnic communities. Simultaneous research focus is also needed to explore parents' experiences of their children's mental health in relation to LLCs.

4.2. Strengths and limitations of the meta-ethnography

4.2.1. Research implications

English language was a selection criterion, and more than 50% of included studies were published in the UK. Parents' experiences of only nine LLCs were examined in these papers. One limitation is that the findings may reflect a cultural bias and a limited healthcare framework. In addition, the age of the children in some studies (fetal alcohol spectrum disorder: 1-37 years, Coons et al. 2016a, b; stroke: 27-46 years, Jones et al. 2012; Juvenile Huntington's disease: 9-24 years, Eatough et al. 2013) may have created a potential recall bias or an inaccurate or incomplete recollection of their parents' experiences when their children were younger. Given the limited number of studies on the above mentioned phenomena, this meta-ethnography has embraced a pragmatic approach. This is in line with Pietkiewicz & Smith's (2012) recommendations regarding group homogeneity in IPA research on rare phenomena or difficult to contact participants. The Juvenile Huntington Disease is a rare condition and at the time of data collection there was no evidence base for

specific interventions (Eatough et al. 2013). People under 65 account for about 25% of those living with stroke, and there is a lack of information regarding young adult stroke survivors (under 65) and their carers (Jones et al. 2012).

4.2.2. Methodological implications

To author's knowledge, this is the first meta-ethnography focusing on parents' experiences of children with LLCs and the first attempt to develop the contribution of IPA by synthesising the findings of available studies in this area. Of particular relevance to this synthesis is the emphasis on the idiographic origin of knowledge. Meta-ethnography is an interpretive form of knowledge that involves a level of abstraction beyond participants' experiences. Although the author recognises that the method added another level of interpretation, the analysis and synthesis processes have been rigorously conducted to allow the conceptual model be grounded in the primary studies. All third- order constructs were checked back against the first- and second- order constructs and the original articles to ensure accuracy and sufficiency. Participants' own words were also used to illustrate some key concepts. All second-order and third-order constructs are presented in Tables 1 and 2 to ensure the transparency of the interpretations. One limitation could be that this synthesis relies on the data presented in each of the included studies which may not reflect the full analysis of the original data. Another limitation refers to the high number of irrelevant papers identified in the literature. This could have been avoided if initially the search focused solely on IPA, thus excluding terms relating to other phenomenological methods or different methods for data collection (e.g. semi-structured, face to face, in-depth).

4.3. Implications for practice

This meta-ethnography indicates that parents valued specialist knowledge and open communication across services. Barriers in communication and lack of professionals'

experience augmented parents' difficulty of understanding their children's illness. Some parents accessed external support or furthered their education to understand the medical language and diagnosis. This is one of the issues reported in a YouGov survey (2016) that indicates that almost 55% of cancer carers in the UK do not receive any support at all. There is a need for a multi-agency and collaborative approach to provide education and training in order to improve parents and clinicians' experience of care (ACT/RCPCH 2009). Some recommendations for practice are highlighted in this meta-ethnography: (1) carers need to be cared for and interventions should fit with the philosophy and values of their family (Brewer et al. 2007); (2) palliative care should include services for carers who experience mental illness relating to their roles, mainly those who feel isolated or stigmatised; (3) equal opportunities for both parents to meet the healthcare team (Ware et al. 2007); (4) end-of-life care discussions should take place prior to a life-threatening episode, as decisions at critical times are difficult and are influenced by emotions (Popejoy 2015). Potential areas of intervention were: *parent-healthcare provider communication, collaborative treatment and care decisions, family intimacy, validation as 'good parent', support to plan goals for future,* and *gender equality in support provision for parents.*

4.4. The need for future research

As identified by other authors (Heinze et al. 2012), currently the majority of studies including parents of children with life-threatening illnesses have been descriptive. Expanding IPA's applicability in more diverse racial, ethnic, socioeconomic, and geographic healthcare contexts would contribute to a broader cultural understanding of service provision. Increasing the use of IPA in the study of a broader LLCs framework would contribute to the development of clinically sensitive interventions. In addition, more IPA research is needed to understand the experiences of caring for a child living with and beyond specific illnesses (e.g.

cystic fibrosis or Juvenile Huntington's Disease), so that parents' care roles and healthcare implications are explored in their unique complexities.

4.5. Some methodological recommendations

No published guidance on how to update a meta-ethnography exists. In order to examine the conceptual development over time of parents' experiences of their children's life-limiting illnesses, three possible approaches can be used: (1) adding to and revising the existing meta-ethnography to incorporate new studies; (2) doing a synthesis of the new studies, then comparing it to the original meta-ethnography; (3) starting the meta-ethnography from the beginning by integrating the older and newer studies to create a single overarching synthesis. The advantage of using the first approach is that the synthesis follows a coherent model, rather than two, and can be efficiently used in practice; there is no arbitrary dividing time between the meta-ethnography in original and update (France et al. 2016). The disadvantage is that the update might be constrained or influenced by the original synthesis, and the process might be challenging for a new team of reviewers if there is no access to the initial findings. The advantage of the second approach is that the analysis process can be innovated by grouping studies according to specific illnesses prior to synthesis.

[table 2 here]

This process can lead to new insights and a more complex model compared to the original meta-ethnography. A potential disadvantage is that the findings from the original and the updated meta-ethnography might not fully integrate, thus making the findings difficult to use in practice. The third approach imposes more effort, however, according to France et al. (2016) it can be used (i) to avoid the updated synthesis being influenced or constrained by the original one; (ii) to avoid having an arbitrary dividing time between the bodies of literature in the original and updated meta-ethnographies; (iii) to redesign the analysis and synthesis process in the update; and/or (iv) to produce a single coherent model that can bring clinical

contribution. Before deciding on which approach to use to update an existing meta-ethnography, it is important to consider the sample of newly available studies. With a high number of papers, starting from the beginning is not feasible. It has been recommended that around 40 studies is the maximum number that can be synthesised efficiently. However, previous syntheses have included over 70 papers (Toye et al. 2014). Computer-assisted coding and extraction of themes may be a way to manage large datasets and help translate studies into one another.

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References

ACT/RCPCH 2009,'*A guide to the development of children's palliative care services'*. 3rd edition. Bristol and London, pp.8-9.

Adams, E, McCann, L, Armes, J, Richardson, A, Stark, D, Watson, E & Hubbard, G 2011, 'The experiences, needs and concerns of younger women with breast cancer: a meta-ethnography', *Psychooncology*, vol. 20, pp. 851–62.

Atkins, S, Lewin, S, Smith, H, Engel, M, Fretheim, A &Volmink, J 2008,'Conducting a meta-ethnography of qualitative literature: lessons learnt', *BMC Med Res Methodol*, 8(21).

Baird, J, Rehm, RS, Hinds, PS, Baggott, C & Davies, B 2016, 'Do You Know My Child? Continuity of Nursing Care in the Pediatric Intensive Care Unit', *Nurs Res*, 65(2), pp. 142–150.

Banning, M 2011, 'Employment and breast cancer: a meta-ethnography', *Eur J Cancer Care*, vol. 20, pp. 708–19.

Bezance, J & Holliday, J 2014 ‘Mothers’ experiences of home treatment for adolescents with anorexia nervosa: an Interpretative Phenomenological Analysis’, *Eating Disorders*, vol. 2, pp 386–404.

Brewer, HM, Smith, JA, Eatough, V, Stanley, CA, Glendinning, NW & Quarrell, OWJ 2007 ‘Caring for a child with Juvenile Huntington’s disease: helpful and unhelpful support’, *Journal of Child Health Care*, vol. 11(1), pp. 40–52.

Burgess, C, Cornelius, V, Love, S, Graham, J, Richards, M & Ramirez, A 2005, ‘Depression and anxiety in women with early breast cancer: five year observational cohort study’, *BMJ*, 330(7493):702.

Burrell, A, Ives, J & Unwin, GJ 2017,’The Experiences of Fathers Who Have Offspring with Autism Spectrum Disorder’, *Journal of Autism and Developmental Disorders*, vol. 47(4), pp 1135–1147.

Campbell, R, Pound, P, Pope, C, Britten, N, Pill, R, Morgan, M & Donovan, J 2003, ‘Evaluating meta-ethnography: a synthesis of qualitative research on lay experience of diabetes and diabetes care’. *Soc Sci Med*, vol. 56, pp. 671–684.

Campbell, R, Pound, P, Morgan, M, Daker-White, G, Britten, N, Pill, R, Yardley, L, Pope, C & Donovan, J 2011, ‘Evaluating metaethnography: systematic analysis and synthesis of qualitative research’, *Health Technol Assess*, vol.15 (43).

Cederborg, AC, Hultman, E & Magnusson, KF 2011, ‘Living with children who have coeliac disease: a parental perspective’, *Child: care, health and development*, vol. 38 (4), pp. 484–489.

Collins, A, Hennessy-Anderson, N, Hosking, S, Hynson, J, Remedios, C & Thomas, K 2016, 'Lived experiences of parents caring for a child with a life-limiting condition in Australia: A qualitative study', *Palliative Medicine*, vol. 30(10), pp. 950–959.

Coons, KD, Watson, SL, Yantzi, NM & Schinke RJ 2016a, 'Adaptation in families raising children with fetal alcohol spectrum disorder. Part II: What would help', *Journal of Intellectual & Developmental Disability*, Vol. 0.

Coons, KD, Watson, SL, Yantzi, NM & Schinke RJ 2016b, 'Adaptation in families raising children with fetal alcohol spectrum disorder. Part II: What would help', *Journal of Intellectual & Developmental Disability*, vol.43 (2).

Cowan, MM 2014, 'The lived experiences of the Sikh population of south east England when caring for a dying relative at home', *International Journal of Palliative Nursing*, vol. 20(4), pp.179-186.

Critical Appraisal Skills Programme 2010, *Critical Appraisal Skills Programme: making sense of evidence about clinical effectiveness: 10 questions to help you make sense of qualitative research*, URL: www.caspuk.net/wpcontent/uploads/2011/11/CASP_Qualitative_Appraisal_Checklist_14oct10.pdf (accessed 24 July 2017).

Dixon-Woods, M, Booth, A & Sutton, A 2007, 'Synthesizing qualitative research: a review of published Reports', *Qual Res*, vol. 7, pp. 375–422.

Eatough, V, Santini, H., Eiser, C, Goller, M-L, Krysa, W, de Nicola, A, Paduanello, Petrollini, M, Rakowicz, M, Squitieri, F, Tibben, A, Weille, KL, Landwehrmeyer, B, Quarrell, O & Smith, JA 2013, 'The personal experience of parenting a child with Juvenile Huntington's Disease: perceptions across Europe', *European Journal of Human Genetics*, vol. 21, pp. 1042–1048.

Elmir, R, Schmied, V & Wilkes, L 2010, 'Women's perceptions and experiences of a traumatic birth: a metaethnography', *J Adv Nurs*, vol. 66, pp. 2142–53.

Forsberg A, Nilsson M, Krantz M & Olausson M 2004, 'The essence of living parental liver donation – donors' lived experiences of donation to their children', *Pediatric Transplantation* vol. 8, pp. 372–380.

France, EF, Wells, M, Lang, H & Williams, B 2016, 'Why, when and how to update a meta-ethnography qualitative synthesis', *Systematic Reviews*, vol. 5 (44).

Garrett, R, Gask, L, Hays, R, Cherrington, A, Bundy, C, Dickens, C, Waheed, W & Coventry, PA 2012, 'Accessing primary health care: a meta-ethnography of the experiences of British South Asian patients with diabetes, coronary heart disease or a mental health problem', *Chronic Illn*, vol. 8(135).

Glasscoe, CA & Smith, JA 2008, 'Through a mother's lens: a qualitative analysis reveals how temporal experience shifts when a boy born preterm has cystic fibrosis', *Clinical Child Psychology and Psychiatry,* vol.13(4), pp. 609-626.

Glasscoe, CA & Smith, JA 2011, 'Unravelling complexities involved in parenting a child with cystic fibrosis: An interpretative phenomenological analysis', *Clinical Child Psychology Psychiatry*, vol.16 (2), pp. 279-298.

Gómez-Ramírez, O, Gibbon, M, Berard, R, Jurencak, R, Green, J, Tucker, L, Shiff, N & Guzman, J 2016,' A recurring rollercoaster ride: a qualitative study of the emotional experiences of parents of children with juvenile idiopathic arthritis', *Pediatr Rheumatol Online J*., vol.14(1):13.

Greenwood, N & Mackenzie, A 2010, 'Informal caring for stroke survivors: meta-ethnographic review of qualitative literature', *Maturitas*, vol. 66, pp. 268–76.

Hain, R, Devins, M, Hastings, R & Noyes, J 2013,'Paediatric palliative care: development and pilot study of a <Directory> of life-limiting conditions'. *BMC Palliative Care*, vol. 12(43).

Hannan, J & Gibson, F 2005, 'Advanced cancer in children: how parents decide on final place of care for their dying child', *International Journal of Palliative Nursing*, vol. 11(6), pp. 284-291.

Hannes, K 2011, 'Critical appraisal of qualitative research'. In: J Noyes, A Booth, K Hannes, A Harden, S Lewin, & C Lockwood, C (eds.) *Supplementary Guidance for Inclusion of Qualitative Research in Cochrane Systematic Reviews of Interventions*, Cochrane Collaboration Qualitative Methods Group, Version 1 Available from: http://cqrmg.cochrane.org/supplemental-handbook-guidance

Hannes, K & Macaitis, K 2012, 'A move to more systematic and transparent approaches in qualitative evidence synthesis: update on a review of published papers'. *Qual Res*, vol. 12, pp. 402–642.

Heinze, E & Nolan, T 2012, 'Parental decision making for children with cancer at the end of life: a metaethnography. *J Pediatr Oncol Nurs*, vol. 29, pp. 337–346.

Hocking, MC, Kazak, AE, Schneider, S, Barkman, D, Barakat, LP & Deatrick, JA 2014,' Parent perspectives on family-based psychosocial interventions in pediatric cancer: a mixed-methods approach', *Support Care Cancer*, vol.22 (5), pp.1287-1294.

Jones, L & Morris, R 2012, 'Experiences of adult stroke survivors and their parent carers: a qualitative study', *Clinical Rehabilitation*, vol. 27(3), pp. 272–280.

Maciver, D, Jones, D & Nicol, M 2010,' Parents' experiences of caring for a child with chronic pain', *Qualitative Health Research*, 20 (9). pp. 1272-1282.

Malpass, A, Shaw, A, Sharp, D, Walter, F, Feder, G, Ridd, M & Kessler, D 2009, '<Medication career> or <moral career>? The two sides of managing anti-depressants: a meta-ethnography of patients' experience of antidepressants', *Soc Sci Med*, vol. 68, pp. 154–68.

Martins, CD, Walker, SP & Fouché, P 2013, 'Fathering a Child with Autism Spectrum Disorder: An Interpretative Phenomenological Analysis, *Indo-Pacific Journal of Phenomenology*, vol. 13(1), pp.1-19.

Mays, N, Pope, C & Popay, J 2005, 'Systematically reviewing qualitative and quantitative evidence to inform management and policy making in the field', *J Health Serv Res Policy* vol. 10, pp. 6–20.

McHugh, ML 2012, 'Interrater reliability: the kappa statistic', *Biochem Med (Zagreb)*, vol. 22(3), pp. 276–282.

NHS England Publications Gateway 2016, *Achieving World-Class Cancer Outcomes: Taking the strategy forward Five Year Forward View*, URL: https://www.england.nhs.uk/wp-content/uploads/2016/05/cancer-strategy.pdf (Accessed April 2018)

Nikfarid, L, Rassouli, M, Borimnejad, L & Alavimajd, H 2017, 'Experience of chronic sorrow in mothers of children with cancer: A phenomenological study', *European Journal of Oncology Nursing*, vol. 28, pp. 98-106.

Noblit, G & Hare, R 1988, *Meta-ethnography: synthesising qualitative studies*. Newbury Park, CA: Sage Publications.

Noyes, J, Hannes, K, Booth, A, Harris, J, Harden, A, Popay, J, Pearson, A, Cargo, M & Pantoja, T 2015, 'Qualitative research and Cochrane reviews'. In: JPT Higgins & S Green (eds.), *Cochrane Handbook for Systematic Reviews of Interventions*, The Cochrane

Collaboration, Version 5.3.0, Available from: http://qim.cochrane.org/supplemental-handbook-guidance (Accessed July 2017)

Pietkiewicz, I & Smith, JA 2012,'Praktyczny przewodnik interpretacyjnej analizy fenomenologicznej w badaniach jakościowych w psychologii', *Czasopismo Psychologiczne*, 18(2), pp.361-369.

Popejoy, E 2015, 'Parents' experiences of care decisions about children with life-limiting illnesses', *Nursing Children and Young People*, vol. 27 (8), pp. 20-24.

Pound, P, Britten, N, Morgan, M, Yardley, L, Pope, C, Daker-White G & Campbell, R 2005, 'Resisting medicines: a synthesis of qualitative studies of medicine taking', *Soc Sci Med*, vol. 61, pp. 133–55.

Randazzo, R, Farmer, K & Lamb, S 2015, 'Queer women's perspectives on sexualization of women in media', *Journal of Bisexuality*, vol. 15, pp. 99–129.

Reilly, DE, Huws, JC, Hastings, RP & Vaughan, FL 2008, 'When your child dies you don't belong in that world any more' – experiences of mothers whose child with an intellectual disability has died', *Journal of Applied Research in Intellectual Disabilities*, vol. 21, pp. 546–560.

Saini, M & Shlonsky, A 2012, *Systematic Synthesis of Qualitative Research*, Oxford: Oxford University Press

Sand, L, Olsson, M & Strang, P 2010, 'What are motives of family members who take responsibility in palliative cancer care?', *Mortality*, vol.15(1), pp. 64-80.

Schweitzer, R, Griffiths, M & Yates. P 2012, 'Parental experience of childhood cancer using Interpretative Phenomenological Analysis', *Psychology and Health*, Vol. 27(6), pp. 704–720.

Seitz, D, Prorok, J & Horgan, S 2012, 'Health care experiences of persons with dementia and their caregivers: a meta-ethnography of qualitative studies', *Alzheimers Dement*, vol. 8(4 Suppl.1):P562.

Smith, JA & Osborn, M 2007, 'Pain as an assault on the self: An interpretative phenomenological analysis of the psychological impact of chronic benign low back pain', *Psychology & Health*, vol. 22(5), pp. 517-534.

Sneeuw, KC, Aaronson, NK, Sprangers, MA, Detmar, SB, Wever, LD & Schornagel, JH 1997, 'Value of caregiver ratings in evaluating the quality of life of patients with cancer', *Journal of Clinical Oncology*, vol. 15(3), pp. 1206-1217.

Thomson, S, Marriott, M, Telford, K, Law, H, McLaughlin, J & Sayal, K 2014, Adolescents with a diagnosis of anorexia nervosa: Parents' experience of recognition and deciding to seek help, *Clinical Child Psychology and Psychiatry*, vol. 19(1), pp. 43–57.

Tong, A, Flemming, K, McInnes, E, Oliver, S & Craig, J 2012, 'Enhancing transparency in reporting the synthesis of qualitative research: ENTREQ'. *BMC Med Res Methodol*, 12:181.

Toye, F, Seers, K, Allcock, N, Briggs, M, Carr, E, Barker, K 2014, 'Meta-ethnography 25 years on: challenges and insights for synthesising a large number of qualitative studies'. *BMC Med Res Methodol*. vol.14 (1):80.

Ware, J & Raval, H 2007, 'A qualitative investigation of fathers' experiences of looking after a child with a life-limiting illness, in process and in retrospect', *Clinical Child Psychology and Psychiatry*, vol. 12(4), pp. 549–565.

Whitehurst, T 2011, 'Raising a child with foetal alcohol syndrome: hearing the parent voice', *British Journal of Learning Disabilities*, vol. 40, pp.187–193.

Wright J, Elwell L, McDonagh JE, Kelly DA & Wray J 2017, 'Parents in transition: experiences of parents of young people with a liver transplant transferring to adult services', *Pediatric Transplant*, vol. 21, e12760.

Xafis, V, Gillam, L, Hynson, J, Sullivan, J, Cossich, M & Wilkinson, D 2015,' Caring Decisions: The Development of a Written Resource for Parents Facing End-of-Life Decisions', *Journal of palliative medicine*, vol.18(11), pp. 945-955.