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Lancaster University Doctorate in Clinical Psychology

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Doctoral Thesis

**Being the Parent of a Child With a Visible Difference: A Focus on Parenting a Child
With a Cleft Lip and/or Palate and Limb Difference**

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Word Count

Thesis Section	Main Text	Appendices (including tables, figures and references)	Total
Thesis Abstract	300		300
Literature Review	7998	8710	16708
Research Paper	7996	9598	17594
Critical Appraisal	3998	642	4640
Ethics	5148	8926	14074
Total	25440	27876	53316

Thesis Abstract

This thesis is primarily concerned with the experience of parenting a child with a visible difference. The focus of the papers were chosen due to their under-representation in the literature and relevance to clinical psychology provision. The thesis commences with a review of the qualitative literature that explored the experience of being a parent of a child with a cleft lip and/or palate. Using Noblit and Hare's (1988) meta-ethnographic approach, 18 papers were reviewed to produce five over-arching themes. The findings illuminated the central experiences associated with the parenting role. Clinical implications concern the enhancement of emotional support for parents within healthcare settings. A pertinent finding emerged relating to the role of parent identity in navigating treatment pathways and its potential influence on treatment decisions.

The research paper explored the experiences of parenting a child with limb difference who has been provided with an artificial limb. Seven parents were interviewed and data analysed using interpretative phenomenological analysis (IPA). Four themes were identified: (1) managing the initial emotional experience through the development of coping resources; (2) opportunities through prosthesis use and its relationship with 'normality'; (3) managing and making sense of social reactions toward their child; (4) the intrinsic role of support: developing a collective connection and enabling shared knowledge. Clinical implications once more concern the provision of emotional support. In addition, the management of social responses and the co-ordination of healthcare services with support networks are discussed. Of relevance to prosthetic rehabilitation services was an interesting divergence related to the experience and meaning of their child's use of a prosthesis concerning functionality.

Finally, the critical appraisal documents the process of reflexivity within the research paper, including the navigation of methodological issues to ensure fidelity with IPA, before discussing the pertinent clinical implications as I move toward qualified life.

Declaration

This thesis presents research submitted in May 2017 as a requirement of the Doctorate in Clinical Psychology at Lancaster University. The research reported is the authors own, except where due reference is made. The research has not been submitted for any other academic award.

Name: James Oliver

Signed:

Date:

Acknowledgements

Firstly, I would like to thank all the participants who so generously gave their time and shared their experiences. I feel privileged to have had the opportunity to speak with you all. I would also like to thank both of my supervisors, Dr Craig Murray and Dr Clare Dixon, for their support and guidance throughout the whole thesis process. It has been a pleasure to work with you both.

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Section One: Literature Review

Being a Parent of a Child with a Cleft Lip and/or Palate: A Qualitative Meta-Synthesis

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Prepared in accordance with the journal guidelines for

Child: Care, Health and Development

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Abstract

Aim: To review and synthesise the available qualitative research, with the aim of developing a conceptual understanding of the experience of being a parent of a child with a cleft lip and/or palate. The review may inform healthcare support from a service user perspective and be of benefit to other parents who have a child with a cleft lip and/or palate.

Method: A systematic literature search was completed utilising five electronic databases. A total of 18 papers met the inclusion and exclusion criteria and were included in the review. Noblit and Hare's (1988) meta-ethnographic approach was used to synthesise the key themes and concepts of the included papers.

Results: The analysis produced five overarching themes: (1) a new idea of parenthood: working through the emotional experience; (2) becoming immersed within a new and unexpected medical system; (3) treatment pathways and the developing parent identity; (4) anticipating and managing the reactions of others; (5) meeting parenting challenges through personal growth and support.

Conclusion: The results highlight central experiences associated with being a parent of a child with a cleft lip and/or palate. Clinical implications are offered concerning the development of parents' self-compassion, and for healthcare professionals to provide effective conditions for emotional containment. A pertinent finding emerged relating to the role of parent identity in navigating treatment pathways and its potential influence on treatment decisions. Future research opportunities are discussed, for instance exploring the experiences of parents with adolescents.

Key Words – child, cleft lip and/or palate, meta-synthesis, parent, qualitative,

Being a Parent of a Child with a Cleft Lip and/or Palate: A Qualitative Meta-Synthesis

A cleft lip and/or palate is a relatively common congenital condition, and occurs in approximately one of every 500–700 births per year globally (World Health Organization, 2012). A definition of the word cleft means to ‘split’ or ‘divide’. During pregnancy, different parts of the face separate before joining together; for a child born with a cleft lip and/or palate, this does not occur. The cause remains largely unknown; however, it is thought to occur through a combination of environmental and genetic factors (Taib, Taib, Swift & van Eeden, 2015).

Antenatal scans, when available, can identify children who will be born with a cleft lip and palate, this usually occurs at the 20-week scan. Children born with solely a cleft palate are unlikely to be diagnosed before birth, with around 1% of cases diagnosed prenatally (Cleft Lip and Palate Association, 2017). The timing of a child’s diagnosis is important to consider, with some parents suggesting a prenatal diagnosis allows them to come to terms with the diagnosis before their child is born (Nusbaum et al., 2008). Supporting parents during this time is a key part of care pathway systems. For instance, in the UK, a referral must be made to a specialist regional cleft network within 24 hours of diagnosis (Taib, et al., 2015). Children born with a cleft lip and/or palate often then engage in a multidisciplinary treatment pathway, which can include involvement with otorhinolaryngology, orthodontics, speech and language therapy, psychology and surgical outcomes (Stock & Feragen, 2016).

Emotional Impact on the Child

There is much research that has explored both the medical and psychosocial impact on children diagnosed with a cleft lip and/or palate. Sharif, Callery and Tierney (2013) refer to a treatment burden for children and their parents: problems with feeding, hearing, speech and language development are common. Surgical intervention is usually completed in the

first year of the child's life to repair the visible features of the cleft, however facial scarring, functional difficulties and speech problems can remain (Stock & Feragen, 2016).

Reviews of the literature focussing on the psychosocial impact of having a cleft lip and/or palate have suggested the majority children do not experience significant difficulties with adjustment or functioning (Hunt, Burden, Hepper & Johnston, 2005; Stock & Feragen, 2016). However, research findings are not consistent, for instance, some studies have found that children are at greater risk of emotional functioning difficulties (e.g., Hunt, Burden, Hepper, Stevenson & Johnston, 2007), whereas other studies have not found this association (Feragen & Stock, 2014). Similarly, some studies suggest children with a cleft lip and/or palate are more likely to have difficulties associated with their social functioning (Slifer et al., 2004), whereas others fail to find this association (Collett, Cloonan, Speltz, Anderka & Werler, 2012). The over reliance on cross-sectional measures and /or retrospective parent, teacher, or healthcare professional reports could contribute to the issue of contradictory findings (Stock & Feragen, 2016). Stock et al. (2016) suggests adjustment to a cleft lip and/or palate is multi-faceted and can fluctuate across time and context, thus making concepts such as adjustment difficult to capture.

Emotional and Social Impact on Parents

There is an abundance of research that has explored parents' responses to their child's diagnosis, with feelings of shock, upset, denial and anger documented (Bradbury & Hewison, 1994; Martin, 2005; Rey-Bellet & Hohlfield, 2004). Often, the birth can lead to parents feeling a sense of guilt; this could be related to parents trying to explore their own role in the cause of their child's cleft condition and blaming themselves (Nelson, O'Leary & Weinman, 2009; Nusbaum et al., 2008).

Studies looking specifically at parents' emotional well-being have highlighted the increased risk of parenting distress during their child's early years (Speltz, Arnsden &

Clarren, 1990). However, by pre-school years, parenting stress levels have often been found to be comparable to parents of children without a cleft lip and/or palate (Berger & Dalton, 2009). Parenting stress is important to consider, with high parental stress found to be a predictor of adjustment difficulties in young children with a cleft condition (Pope, Tillman & Snyder, 2005). This is also a consistent finding within the wider paediatric health literature, with parental adjustment found to be a predictor of child adjustment (Drotar, 1997; Goldberg et al., 1997).

Research has also focussed on the relationship formation between parent and child, informed by attachment theory (Bowlby, 1969). The focus on attachment is in the context of possible disruptions to the parent-child relationship, such as feeding difficulties, hospital admissions and surgeries. However, research suggests that by 2 years of age, children with a cleft lip and/or palate are likely to emerge with secure attachments (Maris, Endriga, Speltze, Jones & Deklyen, 2000; Murray et al., 2008). Research has also indicated that mothers of children with a cleft lip and/or palate can be more encouraging, when compared to mothers of children without a cleft condition (Gassling et al. 2014).

A frequent limitation of the evidence exploring parental distress is the reliance on questionnaire data at fixed timed points. Qualitative studies however, can offer an insight into adjustment over time, with research suggesting several positive aspects of caring for a child with a cleft condition, such as an increased appreciation of diversity, increased acknowledgment of their child's strengths and increased community involvement (Eiserman, 2001; Klein, Pope, Getahun, & Thompson, 2006). Nelson, Glenney, Kirk and Caress (2011) suggest this type of research is important and can help challenge the often negative discourse around 'difference'.

Another key area of research has focussed on the impact of societal views, with Rumsey and Harcourt (2004) suggesting children and their parents' social experiences can be

entangled with feelings of discomfort, anxiety and a fear of rejection. Here, qualitative studies have offered insight into a range of responses. Johansson and Ringsberg (2004) found parents worried about the reactions of others, including stares, a look of fear, and negative comments aimed at their child. In addition, parents have reported worrying about future social issues including social acceptance (Klein et al., 2006), and experience emotional distress when they hear of their child being teased or rejected (Klein, Pope & Abbott, 2014). Tiemens, Nicholas and Forrest (2013) suggest close relationships with families and friends can act as a shield against some of the social challenges children with a cleft lip and/or palate can encounter.

Parents' Experiences of Services

In recent years, there has been a growing interest in parents' experiences of healthcare services. Several quantitative studies have explored aspects of healthcare support. Young, O'Riordan, Goldstein and Robin (2001), used self-administered questionnaires and found that parents' priorities were related to feeding, the identification of health issues, and a desire to be shown the normal aspects of their baby's examination. Furthermore, a large percentage of parents wanted to know about the aetiology and be reassured regarding their own role in the diagnosis of their child's cleft lip and/or palate. Many parents felt these needs were not addressed at the time of the birth of their baby. Byrnes, Berk, Cooper and Marazita (2003), once more using self-administered questionnaires, found parents wanted the informing healthcare professionals to be in greater control of the conversation. In addition, they wanted clinicians to show more of their own feelings, and give parents the opportunity to discuss their emotional responses. Furthermore, they found a positive association between parent satisfaction and the degree to which they felt they knew the healthcare professional, highlighting the importance of the perceived relationship.

Nelson and Kirk (2013) completed a qualitative study exploring parent experiences of services, which enabled a detailed insight into valued aspects of care. They found positive appraisals to be founded on attributions of cleft practitioners as competent and trustworthy, displaying highly developed communication skills and offering continuity of care. Areas of unmet need centred around individualised information regarding treatment, and the co-ordination of services across the course of their child's treatment.

Rationale

To summarise, the research literature suggests that being the parent of a child with a cleft lip and/or palate can be both a rewarding and challenging role. Much of the research has used a quantitative design, which has enabled several important insights into the experiences of parents to be gained, including the experience of diagnosis and the psychosocial impact. Exploring the experiences of parents is of significant value, not least because of the research highlighting the association between parental adjustment and child adjustment (Pope et al., 2005).

Nelson et al. (2011) suggest much of the quantitative literature exploring parent experiences has been conducted with small sample sizes, using cross-sectional designs focused mainly on mothers. This can produce a static picture of variables at fixed time points and contexts, making generalisations difficult and findings which lack depth. Qualitative research on the other hand, view experiences in flux and offer scope for a greater range of emotional responses, as it is based on data generation and sensitive to social context (Mason, 2002). Nelson (2009) suggests qualitative research within the craniofacial field can shed light into the complexity of social life, and provide insight to beliefs and behaviour. Interestingly, Stock and Feragen (2016) note that research within the field of cleft lip and/or palate has seen a recent shift toward a more balanced, inclusive and patient-driven approach. However, to

date, there is no published qualitative meta-synthesis that has integrated the qualitative research findings.

Walsh and Downe (2005) suggests a qualitative meta-synthesis allows for the amalgamation of findings from qualitative research on a given topic area and seeks to understand and explain phenomenon. The utilisation of qualitative meta-syntheses are being increasingly used within healthcare research, and can facilitate shared knowledge and inform policy and practice (Dixon-Woods, Booth, & Sutton, 2007; Toye et al., 2014). Schreiber, Crooks and Stern (1997) described the approach as the “bringing together and breaking down of findings, examining them, discovering the essential features, and, in some way, combining phenomena into a transformed whole” (p.314). By reviewing the research that has prioritised the lived experiences of parents, it may help shed light into how parents experience their parenting role, the meanings ascribed to these experiences, and offer an insight into a range of emotional responses, beliefs and coping resources. A review of this nature seems particularly poignant given the increasing involvement of service-users influencing the design and structure of their own healthcare experience (Crawford et al., 2002). The research aim is therefore to review and synthesise the qualitative research exploring the experience of being a parent of a child with a cleft lip and/or palate. The research question is “What is the experience of parenting a child with a cleft lip and/or palate and how do parents make sense of the parenting role?”

Method

Noblit and Hare’s (1988) meta-ethnographic approach was used to conduct the meta-synthesis. The aim was to produce an interpretative analysis of the studies exploring the experience and meaning of parenting of a child with a cleft lip and/or palate. The approach looks to produce third-order interpretations of experiences; that is, it looks to synthesise in an integrative thematic structure, the authors’ interpretations (second-order constructs) of the

participant's understandings of their own experiences (first-order constructs) as reported in the reviewed papers (Atkins et al. 2008).

Search Strategy & Selection Strategy

Studies included in the review were identified after completing a systematic search of the bibliographic databases MEDLINE (covering biomedical and health related literature; searchable years 1806-2016), PsychINFO (covering behavioural and social science literature; searchable years 1806-2016), CINAHL (covering nursing and allied health disciplines; searchable years 1937-2016), Web of Science (covering art, humanity and social science literature; searchable years 1900-2016) and AMED (covering complimentary medicine and allied professional literature; searchable years 1985-2016). The research question was broken down into four distinct concepts to help construct an effective and comprehensive search strategy. In addition, both a specialist university librarian and a local clinician working with children with a cleft lip and/or palate were consulted for feedback regarding the search terms and strategy. The strategy consisted of utilising the respective database subject headings with a range of free text search terms. Searches were limited to title and abstract. The boolean operator 'OR' was used to search for terms within each concept, with the boolean operator 'AND' used between concepts. Appendix A illustrates the search terms and strategy utilised for each bibliographic database.

To explore the research question in the most defined manner, inclusion and exclusion criteria were applied to the studies included in the review. The inclusion criteria were as follows: (1) the study was written in English, (2) the study was published in a peer-reviewed journal (to ensure a minimum level of quality appraisal), (3) the study utilised 1:1 interviews or focus groups for data collection and an inductive qualitative approach to analyse the data, (4) the sample consisted of parents or caregivers of children with a cleft lip and/or palate, (5) the study explored first person accounts of parents or caregivers, evidenced by original data

excerpts, (6) the authors provided interpretations of parents' experiences based on the qualitative data obtained, (7) the study included findings relating to the experience of parenting a child with a cleft lip and/or palate and how it is to be a parent of a child with a cleft lip and/or palate.

Conversely, studies were excluded if they (1) focused on parents' evaluations of, or satisfaction with, services unless the themes produced related to how it is to be a parent of a child with a cleft lip and/or palate, or (2) they focussed on parenting experiences of other physical health conditions as well as a diagnosis of a cleft lip and/or palate.

The search was completed on 21st November 2016 and produced a total of 1161 search results. The results were collated into a referencing software program, before being visually scanned by title and abstract to assess suitability for inclusion in the review. Where relevance was deemed ambiguous, the full text version of the paper was accessed. Finally, the reference lists of the included papers were visually scanned to check for additional papers; this yielded one additional paper. A total of eighteen papers were identified as being pertinent to the research question and included in the review. Figure 1 illustrates the systematic search process.

[INSERT FIGURE 1]

Characteristics of the Selected Studies

Eighteen papers were identified for the meta-synthesis. All papers were published between 2004 and 2016. Eight papers used European samples (6 in the UK, 1 in Norway, 1 in Sweden), seven papers used samples in the USA, two used a sample in Brazil, and one paper used a sample in Taiwan. Sample sizes ranged from 8 to 118 and included birth mothers' and fathers' perspectives. Seventeen papers used one-to-one interviews with parents for data collection, and one paper utilised a focus group design. Two papers used the same sample to

generate the research findings (Nelson, Caress, et al., 2012; Nelson, Kirk, et al., 2012); these papers were included as they met the inclusion criteria and reported different aspects of the analysed data in the identification of themes.

All the included papers used some form of an inductive thematic analysis. Specifically, twelve papers used variations of a phenomenological thematic method, four used a grounded theory approach and two used an unspecified generic thematic analysis. Two papers used a mixed method design, with only the qualitative analysis extracted in the synthesis. All the studies can be claimed to fit within a critical realist framework; which acknowledges that the language used by participants to be an understanding of their ‘truth’ and reflect their thoughts, beliefs and experiences; thereby making it possible to understand their perspective (Maxwell, 2012). Table 1 highlights the key characteristics of the included studies. The varied nature of the samples within the included papers can be considered a strength of the review in that it presents a balance between a homogenous sample within a heterogeneous sample context. This allowed insight into the robustness of findings across sample contexts and how experiences varied within themes.

[INSERT TABLE 1]

Quality Appraisal of the Selected Studies

The Critical Appraisal Skills Programme (CASP; Public Health Resource Unit, 2006) was utilised as a framework for assessing the quality of the included papers. This framework invites the reviewer to consider ten questions that can help guide decisions regarding quality. These questions include the appropriateness of the research design and recruitment, the rigor of the analysis and the value of the research. To assess quality, the review adopted a scoring system used by Duggleby et al. (2010), which assesses quality on a 3-point scoring system. A score of 1 represents a weak explanation, a score of 2 represents a moderate report, and a

score of 3 represents a strong report and justification of the particular issue. CASP scores ranged from 13 to 24 (appendix B).

No papers were excluded on the basis of reviewed quality, with debates ongoing around what constitutes high quality qualitative research (Barbour, 2001; Ring, Jepson & Ritchie, 2011). The use of checklists have helped convince some sceptics of the rigour and robustness of qualitative research and appear to be endorsed by guidance by the Centre for Reviews and Dissemination (2009). However, often the key indicator of quality can be a description detailed enough for the reader to make an informed judgement (Popay, Rodgers & Williams, 1998). Furthermore, complex dilemmas regarding qualitative design cannot always be solved by formulaic checklists (Barbour, 2001). The present review is therefore adopting the approach of Sandelowski, Docherty and Emden's (1997) argument that "studies should not be excluded for reasons of quality, because, as we noted previously, there are wide variations in conceptions of the good, and in quality criteria" (p.368). Within the present review its inclusion can help to guide the reader regarding the reported quality of the included research, and offer assurance that the themes presented within the review were not predicated on studies with lower CASP scores.

Analysing and Synthesizing the Selected Studies

Data extraction and analysis took place in the following way. Included studies were firstly read several times to become familiar with the data set. Relationships between studies were sought with the listing of key themes and concepts from each paper that were relevant to the research question. The notion of first order and second-order constructs were helpful at this stage, with the author's second-order constructs (their interpretations of the participant's experiences) extracted, along with participant quotes or phrases (first-order constructs) to ensure subsequent interpretations were grounded within the data. These interpretations were

then compared across studies and into discrete concepts to produce emerging third-order interpretations. Through continued iteration it was possible to identify similarities and differences between concepts and organise them into final third-order themes. For example, the theme 'Meeting Parenting Challenges Through Personal Growth and Support' came from frequently reported aspects of coping and resilience, as well as the value parents placed on support to meet the challenges of the role. It is these third-order themes that illustrate the interpretative nature of the review and looks to extend the meaning of the reviewed papers. Noblit and Hare (1988) define the synthesis as "making a whole into something more than the parts alone imply" (p.28). Appendices C to E illustrate this audit trail.

Results

The analysis led to the generation of five main overarching themes. They are discussed in detail below:

Theme 1. A New Idea of Parenthood: Working Through the Emotional Experience

Adjusting to a new idea of being a parent and the emotional experience this entails was detailed within thirteen of the included papers (Hsieh et al., 2013; Johansson & Ringsberg, 2004; Lindberg & Berglund, 2014; Martins et al., 2013; McCorkell et al., 2012; Nelson, Kirk, et al., 2012; Nusbaum et al., 2008; Stock & Rumsey, 2015; Stone et al., 2010; Tierney et al., 2015; Vanz & Ribeiro, 2011; Williams et al., 2012; Zeytinoglu et al., 2016). The emotions the authors reported included shock, worry, despair, sadness, denial, shame, and guilt, but also positive emotions such as happiness: "My first reaction was shock. I wasn't really prepared. We didn't think we'd have a child with a cleft palate. It was a funny feeling. I thought he was ugly but sweet at the same time" (Johansson & Ringsberg, 2004, p.167).

A powerful emotional response came through the analysis, relating to letting go of the parenting assumptions they previously held and the idea of a 'perfect' child. One parent reported:

It's very difficult to take in that the child you thought was "perfect" in inverted commas, turns out that they're not perfect as defined by medical professionals. I understand why she's described as abnormal, but to me she was perfect, she wasn't abnormal. (Nelson, Kirk, et al., 2012, p.350)

In Nusbaum et al.'s (2008) paper, the authors report how one parent discussed the grieving process they went through when they realised their baby wasn't going to be "perfect": "I think the most important [thing] is like, you do your mourning...When someone dies, you mourn for a while. So doing that before really helps..." (p.342).

Related to this notion of adjusting to a new idea of parenthood were a number of powerful emotions concerning parent's desire to provide for their child. Concerns relating to breastfeeding were a particularly strong theme: "It was overwhelming because all thoughts came at the same time...what will she look like...how can I feed her" (Lindberg & Berglund, 2014 p.68). One parent reported feelings of guilt in relation to being unable to provide a "perfect body for their child" (Stone et al., 2010, p.1359). Another parent reported a sense of determination when seeing her child for the first time: "I saw it just before they picked him up and laid him on his stomach. That's when I saw it. I was really shocked but I thought, 'Well, he's chosen the right parents'" (Johansson & Ringsberg, 2004, p.167).

The timing of the child's diagnosis did not seem to have an impact on the range of emotions expressed, however there were several themes that suggested a diagnosis prenatally enabled a period of preparation: "It was very beneficial to know prior to birth...The part of it being a shock was over, it was dealt with prior to birth" (Nusbaum et al., 2008, p.342).

From the synthesis, there was a sense of understanding to parents' emotional experience and something they needed time to experience and work through as they adjusted to a new idea of parenthood.

Theme 2. Becoming Immersed Within a New and Unexpected Medical System

The experience of entering a new and unexpected medical system and its interaction with parents' emotional experience, particularly their perceived ability to cope and adjust to their new role, was detailed within seventeen of the included papers. (Hopkins et al., 2016; Hsieh et al., 2013; Johansson & Ringsberg, 2004; Lindberg & Berglund, 2014; Martins et al., 2013; McCorkell et al., 2012; Nelson, Caress, et al., 2012; Nelson, Kirk, et al., 2012; Nusbaum et al., 2008; Shipe et al., 2016; Sischo et al., 2015; Sischo et al., 2016; Stock & Rumsey, 2015; Stone et al., 2010; Tierney et al., 2015; Vanz & Ribeiro, 2011; Williams et al., 2012).

Parents appeared to look for support or emotional containment from those in a position of professional power. One parent recalled the following experience: "My partner was crying her eyes out and we were left alone in this room. Then a staff nurse put a piece of paper in front of us and I will always remember it said 'how to deal with a disabled child'" (Stock & Rumsey, 2015, p.33). Here, emotional containment was lacking and in fact resulted in increased distress for parents. Tierney et al. (2015) reported parents seeking a late diagnosis felt like their parental instincts were being ignored by healthcare professionals. Similarly, Johansson and Ringsberg (2004) suggested attempts by staff to contain parents' emotions, while good intentioned, could have the opposite effect: "He will be fine later on" (p.168), giving the implicit message that their child was not fine now.

For some parents the experience of being confronted with the treatment pathway could be overwhelming. "I say that the work with them, I think, never ends, [I] even

mentioned it to my husband, I think it's a continuous thing... We go and there is always something new" (Martins et al., 2013, p. 496). Some parents compared future treatment outcomes to a "guessing game" (Nelson, Kirk, et al., 2012, p.352), highlighting the sense of uncertainty they experienced.

From the synthesis, perceived professional knowledge was an important factor in feeling supported, and feeling capable of meeting the challenges of their parenting role. Lindberg and Berglund (2014) reported on the impact of receiving contradictory information which could unsettle parent confidence in their ability to provide for their child. Similarly, Vanz and Ribeiro (2011) suggested vague information could leave parents feeling insecure. Perceived professional knowledge seemed to reduce parental feelings of disquiet, and reduce a burdening sense of responsibility to choose the correct treatment: "At the end of the day if he [orthodontist] thinks that's the right thing, then who am I to judge it...if they think it's the right way to go, then who are we to disagree?" (Nelson, Caress, et al., 2012, p.801).

A responsive, caring and knowledgeable healthcare team was valued by many parents and seemed to provide a sense of trust and emotional containment during what was for many parents a turbulent emotional period: "My midwife was fantastic. She was perfectly calm, [and] pointed out that he was strong and healthy. She made me realise I should be thankful for that" (Johansson & Ringsberg, 2004, p. 168); "The staff was amazing...they're very therapeutic and loving" (Hopkins et al., 2016, p.2418).

Interestingly, from the synthesis there did seem to be a discrepancy between perceived support from general healthcare professionals and specialist cleft teams, with the latter generally receiving positive comments when discussed. Nelson, Caress, et al. (2012) suggest parents construct a sense of 'being in the "right" hands', and trust the specialists providing cleft care (p.800). Another parent referred to the specialist cleft team as an

“extended family” (Sischo et al., 2016, p.479); therefore, affirming the importance of parents feeling supported during this vulnerable time as they adjust to a new idea of being a parent, and enter a new and unexpected medical system.

Theme 3. Treatment Pathways and the Developing Parent Identity

This theme encapsulates how parents’ developing sense of identity seemed to shape their involvement in the treatment pathway and the decisions they made for their child; it was detailed within ten of the reviewed papers (Hopkins et al., 2016; Hsieh et al., 2013; Johansson & Ringsberg, 2004; Lindberg & Berglund, 2014; Nelson, Caress, et al., 2012; Nelson, Kirk, et al., 2012; Shipe et al., 2016; Sischo et al., 2015; Sischo et al., 2016; Zeytinoglu et al., 2016).

Some parents reported acting or taking decisions on behalf of their children in the context of doing the best for their child: “When it is your child you just want the best for them don’t you, when you’re a parent?” (Nelson, Caress, et al. 2012, p.798). Nelson, Caress, et al. (2012) reported a moral aspect of decisions around surgery, with parents feeling a need to ‘do the right thing’. This could contribute to a perceived obligation to accept all treatment options on offer, in a sense show they are virtuous and responsible parents and ultimately helping their child. Similarly, Hopkins et al. (2016) reported that parents using a procedure known as nasoalveolar moulding (NAM), a nonsurgical treatment option involving an intensive regimen for parents, showed a strong sense of desire and determination to give their child the best care: “It was rough going up (to the clinic in another city) once a week and trying to balance work and everything, I will say that. But I think that you do what you have to do for your kids and you make it work” (p.2417).

Some parents had conflicting emotions when making decisions regarding their child’s surgery, with lip repair usually considered in children as young as three months old. It could

be difficult for parents to reduce the tension between a surgery decision that may improve a child's function or 'normalise' appearance, and their parental instincts to protect their child: "At the end of the day, we knew at the back of our minds we needed to do something... what your heart tells you and your head tells you are two different things?" (Nelson, Kirk, et al., 2012, p.350). Some parents seemed to view surgeries as a necessity: "You know it's not an option not to get this done" (Stock & Rumsey, 2015, p.36).

Interestingly, in Shipe et al.'s (2016) paper, which looked at parents of internationally adopted children with a cleft lip and/or palate, attachment was described as an aspect of decision making in relation to their child's surgery decision. Some parents worried about the relationship and bond with their child, which seemed to be exacerbated by language differences between parent and child: "We weren't that attached yet, and how were we going to comfort this child we couldn't communicate with?" (p.447). The explicit focus on attachment as a consideration in surgery decisions seems to be particularly poignant to adopted parents.

Theme 4. Anticipating and Managing the Reactions of Others

Parent reports of worries or concerns relating to the reactions of other people was expressed in ten of the reviewed papers (Hsieh et al., 2013; Johansson & Ringsberg, 2004; Nelson, Caress, et al., 2012; Nelson, Kirk, et al., 2012; Nusbaum et al., 2008; Sischo et al., 2016; Stock & Rumsey, 2015; Stone et al., 2010; Vanz & Ribeiro, 2011; Zeytinoğlu et al., 2016). Many parents reported being concerned about the reaction from friends and family, however mostly these concerns were unfounded: "My mum came to the maternity ward. She picked him up and held him and established a very special relationship with him right from the start. She said he was lovely and had such a beautiful head" (Johansson & Ringsberg, 2004, p.169). Many parents took great comfort from their family and friends' support.

However, a lack of support could be distressing for parents: “I got comments like ‘Well, it’s not from our side of the family’...I was almost having to absorb their grief, instead of them saying, ‘What can I do to help?’” (Stock & Rumsey, 2015, p.35).

Reactions from the public were also an ongoing concern for many parents, with reported instances of negative reactions: “You’d walk past people and they used to look and say ‘oh can I have a look at your baby?’ What used to upset me was hearing them talking behind your back as you’re going away” (Nelson, Kirk, et al., 2012, p.352).

Anticipatory concerns relating to societal reactions were prominent parental experiences, with concerns relating to their child’s future: “The day she comes home telling me she’s being teased will break my heart, but there is nothing we can do about it” (Stock & Rumsey, 2015, p.37). On closer inspection, many of these concerns seemed to relate to parents’ own views and assumptions relating to difference, more specifically, how society responds to visible difference. Parents seemed to worry about the impact of their child looking or sounding different: “He needs to be looking like the rest of them [at college] to be able to socialise with the rest of them” (Nelson, Caress, et al., 2012, p.800). Another parent added: “I’m keen that his cleft doesn’t define him or interfere with his childhood...” (Stock & Rumsey, 2015, p.37).

Theme 5. Meeting Parenting Challenges Through Personal Growth and Support

The final theme was detailed in ten of the reviewed papers and relates to the personal growth parents reported, and the support they relied on to cope (Hopkins et al., 2016; Lindberg & Berglund, 2014; Martins et al., 2013; Nusbaum et al., 2008; Sischo et al., 2015; Sischo et al., 2016; Stock & Rumsey, 2015; Stone et al., 2010; Williams et al., 2012; Zeytinoğlu et al., 2016).

Parents seemed to have an increased appreciation of their own resourcefulness and ability to adapt: “Having babies before, it was a lot like having blinkers on...this opens your eyes to what else is out there. We’re learning all the time” (Stock & Rumsey, 2015, p.37). Another parent reflected: “About myself, I guess, I’ve learned that I’m capable of just doing whatever I have to do” (Sischo et al., 2015, p.11). Some parents reflected that they had learnt to live one day at a time and maintain a positive outlook: “The most important thing is just to keep a positive attitude” (Sischo et al., 2016, p. 479).

Support was commonly discussed as a way of managing the challenges parents were presented with. Working together with a partner, and sharing the emotional and practical load, was as an important source of support: “He has been very good in supporting” (Lindberg & Berglund, 2014, p.70). Support from other parents of children with a cleft lip and/or palate was also highly valued: “It was very important to know that it wasn’t just my reality what I was going through, that there were other people going through this and helping me...” (Martins et al., 2013, p.497). Seeking support from other parents could also interact with personal growth and help contain some of the anxieties parents often encounter: The role of peer support appears to be encapsulated by the following parent:

I have found that shared experiences are the most valuable and could help dispel a majority of the unknowns and anxiety. There are also so many things that could be shared to aid in caring for the special needs of these children. (Nusbaum et al., 2008, p.341)

The value parents placed on the support they received seems to be epitomised by a desire to reciprocate and look to support other parents (Hopkins et al., 2016; Martins et al., 2013; Stock & Rumsey, 2015): “We now offer our support and experiences to expecting

parents...because we've been through it, we're trying to reach out to help people" (Stock & Rumsey, 2015, p.37).

Discussion

The review has synthesised the experiences of being a parent of a child with a cleft lip and/or palate, and in doing so, illustrated key aspects of their parenting experience. The review suggests parents go through a period of adjustment as they come to terms with a new idea of being a parent. The application of the meta-synthesis methodology has enabled new insights to be achieved with key concepts and themes gathered across a wide body of literature. The literature varies according to sample, country and cultural context; but importantly remains homogenous by including only parents of children with a cleft lip and/or palate.

The results of the review suggest being the parent of a child with a cleft lip and/or palate includes a period of adjustment to a new idea of parenthood. Many strong emotions were expressed, and there was a sense that parents must work through these emotions as they adapt to a new idea of parenthood. A novel aspect within the theme was the idea that some of these emotional responses seemed to be linked to doubts regarding their ability to provide for their child. Parent experiences relating to the loss of a 'perfect' child was also expressed, this idea has been discussed previously in the research literature (Fajardo, 1987).

The review suggests the immersion within a medical system is a central aspect of being a parent of a child with a cleft lip and/or palate and can influence parents' emotional experience. Parents reported that the manner of healthcare professional's communication, particularly at the time of diagnosis or birth of their child, could exacerbate feelings of uncertainty and anxiety. This finding has received some attention in the past with Searle, Ryan and Waylen (2016), who found discontent amongst parents' evaluation of healthcare

professional's communication when diagnosing their child with a cleft lip and/or palate. The review extends this finding and suggests emotional containment by healthcare professionals and perceived competence are important aspects of support satisfaction. There are good practice guidelines for healthcare professionals in discussing difficult health news with parents (e.g. SCOPE, 2003), but given parents frequently expressed dissatisfaction around this aspect of support, perhaps a greater emphasis on training around the psychological impact of a child's diagnosis on parents should be promoted.

A novel finding within the review was the interaction between treatment decisions and the identity of being a parent, particularly in parents wanting to do all they can for their child. These treatment decisions are founded within a heightened emotional framework, and seemed to create a tension between a need to protect their child but also accept all available treatment options. This tension is important for healthcare professionals to be aware of, particularly within the context of collaborative decision making. In the UK for example, the NHS constitution for England (Department of Health, 2015) applies the concept on patient-centred care, with a central feature of this being a collaborative approach to decision making. The present theme suggests care needs to be taken with this assumption of collaborative decision making, as decisions relating to surgery may be driven by an unspoken, and possibly an unconscious need, to be the best parents and do all they can for their child.

A prominent aspect of parent experience within the present review was the anticipation and management of social reactions, and worrying how others will treat their child. This aspect of parent experience has been previously reported (Klein et al., 2006), and could link to wider societal ideas and views related to visible difference. Wardle and Boyce (2009) analysed the content of television programming and found people with a visible difference were rarely shown on television and when they did would often be cast in roles with a negative stereotype. This highlights the paradox of visible difference often being

'invisible' in mainstream media. The role of charities such as Changing Faces in the UK, and its campaigns such as *Face Equality* (Changing Faces, 2017), are paramount in continuing to challenge discrimination and prejudice around issues related to visible difference.

The final theme suggests the experience of being a parent, and overcoming several stressors and challenges, can bring a sense of personal growth. Parents discussed the importance of parent-to-parent support in providing succour in times of challenge. Trenchard et al. (2014) evaluated an antenatal group for parents expecting a baby with a cleft lip and/or palate using thematic analysis of open ended questionnaire data. They found themes relating to the benefits of meeting other parents, including their worries being normalised, and feeling a sense of connection and reassurance. Parents also found the group allowed them space to focus on the joys and excitement associated with the birth of their child. The importance of parent support may link to the development of resilience. Resilience can be described as the successful adaptation to significant adversity, using protective factors to respond to change (Beardslee, Versage & Gladstone, 1998). This description is important to consider, as parent growth was closely related to peer support and appeared to help parents overcome the challenges inherent within the role.

Clinical Implications

The findings from the review can have several important clinical implications to help support parents. Supporting parents to manage the powerful emotions they report such as shock, despair, shame and guilt is paramount. Some of these feelings seemed to relate to parents doubting their own ability to be able to provide for their child. Compassion-focussed therapy has been shown to help support individuals with feelings of shame, guilt and self-criticism (Gilbert, 2009). It may help support parents who are experiencing these strong emotions in relation to feeling that they have let their child down in some way, for example, a

feeling of guilt regarding a possible genetic influence, or concerns such as breastfeeding difficulties. Supporting the development of self-compassion may also support parents to become more aware of any moral drivers to decisions around surgery, and feel less of a need to prove they are virtuous parents.

The review suggests a sense of feeling supported within healthcare systems and emotionally contained was an important aspect of parent experience. The relationship between parent and healthcare provider has received a lot of attention in the research literature, with positive parent-physician and child-physician relationships associated with satisfaction of care with paediatric settings (Swedlund, Schumacher, Young & Cox, 2012) and specialist cleft services (Nelson & Kirk, 2013). Thinking about the relationship more broadly, in the UK there is increasing attention given to the development of clinical services, specifically mental health services, using the principles of attachment theory (Bowlby, 1969). This involves thinking carefully about service user experience in a relationally informed lens. Advocates want clinical services to become more psychologically conversant and attachment informed (Bucci, Roberts, Danquah & Berry, 2015, Seager, 2014). For instance, in Bucci et al.'s (2015) review, this involved services thinking about attachment in all aspects of service design and delivery, starting from the referral pathway, and services ensuring that there will be at least one stable relationship with a professional. Papers within the review discussed the importance of staff sensitivity to client distress, and the significance of human contact and comfort (e.g. Goodwin, Holmes, Cochrane & Mason., 2003). Thinking about attachment needs through service design could also be considered within paediatric health settings, with parents in the present review valuing sensitive support, reassurance and the chance to talk through their emotions; particularly from specialist cleft teams.

Trust was reported by parents as being an important aspect of feeling supported. Interestingly, Nelson, Caress, et al. (2012) found at times, this trust could be fatalistic, and

appeal to a surgical heroism, whereby the risks or consequences of surgery could be downplayed by parents to protect the trust they placed in their healthcare team. Thinking about professional power here is important, and perhaps healthcare professionals have a duty of care to look at ways of reducing the inherent power imbalances within parent-provider relationships. This may offer a role for psychologists, in particular team formulation, to highlight the role of professional power more explicitly within parent-provider relationships. Team formulation refers to the process of facilitating a team of professionals to construct a shared understanding of a difficulty or dilemma; this could involve decisions or discussions around surgery options. Its use can be an effective means to shift multidisciplinary team cultures toward psychosocial perspectives (Division of Clinical Psychology, 2011). This suggestion would also fit with the guidance offered by the British Psychological Society's (2007) *New Ways of Working for Applied Psychologists in Health and Social Care: Working Psychologically in Teams*, which recommends psychologists promote the unique contribution of their work within healthcare teams.

The review also highlighted the role of systemic influences on parents, particularly the role of social responses. Family systems theories can offer a helpful framework to explore systemic influences (Bowen, 1978; Minuchin, 1974). The theories look to view the family as one unit with problems and difficulties framed within a family system, rather than within the individual. In the context of the present review, it may help parents to explore trans-generational issues, and their own relationship with bullying, teasing and 'difference'. This may unconsciously influence how they anticipate and respond to negative social experiences and model responses to their child. Thinking carefully about these matters with a trained mental health professional may offer helpful clinical support in managing the reaction of others.

Limitations

The review does have several limitations. Firstly, and consistent with parenting research more broadly, fathers' experiences were under-represented (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005). Two of the included papers were focussed on father's experiences (Stock & Rumsey, 2015; Zeytinoğlu et al., 2016) which is a positive step. Findings from these studies showed that many fathers take an active role in parenting their child and experience similar emotional responses as mothers. However, further efforts to address this discrepancy should be prioritised. Secondly, the reviewed papers were mainly focussed on parents' experiences relating to young children. A broader range of ages may bring into focus different aspects of parent experience. A final limitation concerns a broader methodological issue: the influence of the author in the synthesis of the studies. A meta-ethnographic approach is interpretative in nature, therefore viewing this as a limitation may well be fallacious, with the aim to develop a third-order interpretative account. As Yardley (2008) comments, researchers within the qualitative research paradigm accept the inevitable influence they have on the research process, with reflexivity considered throughout. The generation of a throughout audit trail (appendices C to E) offers assurances to the reader regarding the credibility and validity of the analysis.

Future Research

The review has highlighted several opportunities for future research. Further research exploring the experiences of parenting adolescent children and their transition to adulthood would be of value. Erikson's (1968) psychosocial stage model of development reports adolescence as a key time, and is associated with an identity and role confusion conflict. Additionally, there is an increasing interest in the aetiology of a cleft lip and/or palate. The present review uncovered strong emotions such as guilt as parents looked to understand their own role in their child's diagnosis. Further research may build on this and compare the

experiences of parents who have been found to have a possible genetic influence with those who have not, and think about effective support for these parents.

Future research should also continue to make concerted efforts to engage fathers in paediatric research. Research into parenting experiences may currently be neglecting meaningful differences related to the experiences of fathers. Phares et al. (2005) offer a thoughtful discussion around overcoming some of the issues in the current under-representation of father's voices within paediatric research. They offer suggestions relating to research design, recruitment and the analysis of data. Purposeful efforts to engage with some of these considerations to include fathers' voices may help to enhance the generalisability of findings within parenting research.

Finally, exploring the development of resilience and the conditions that enable parental adaptation would be beneficial. Baker, Owens, Stern and Willmot's (2009) cross-sectional study found parental coping style and perceived social support to be related to positive adjustment in parents of a child with a cleft lip and/or palate: research could explore this further using a complimentary qualitative approach.

Conclusion

The present review explored the experience of being a parent of a child with a cleft lip and/or palate. The review successfully synthesised studies from a variety of countries, cultures and contexts, and highlighted five over-arching themes that encapsulate how it is to be a parent. The review suggests that parents experience a variety of emotions as they adjust to a new idea of parenthood. The experience of being immersed within a new and unexpected medical system is a central feature of the experience, with a novel finding around parent identity and treatment decisions emerging from the synthesis. The review suggests parents contend with social challenges, particularly the reaction of others, and seem to develop a

sense of growth as they adjust to being a parent of a child with a cleft lip and/or palate. There are several implications for healthcare services and professionals, including a greater role for emotional support, consideration of professional power, and a greater priority in fostering relationships with parents.

References

*indicates paper included in review

Atkins, S., Lewin, S., Smith, H., Engel, M., Fretheim, A., & Volmink, J. (2008). Conducting a meta-ethnography of qualitative literature: lessons learnt. *BMC Medical Research Methodology*, 821. doi:10.1186/1471-2288-8-21

Baker, S. R., Owens, J., Stern, M., & Willmot, D. (2009). Coping strategies and social support in the family impact of cleft lip and palate and parents' adjustment and psychological distress. *The Cleft Palate-Craniofacial Journal*, 46(3), 229-236. doi:10.1597/08-075.1

Barbour, R. S. (2001). Checklists for improving rigour in qualitative research: a case of the tail wagging the dog? *BMJ (Clinical Research Ed.)*, 322(7294), 1115-1117. doi:10.1136/bmj.322.7294.1115

Beardslee, W. R., Versage, E. M., & Gladstone, T. R. (1998). Children of affectively ill parents: a review of the past 10 years. *Journal of The American Academy of Child and Adolescent Psychiatry*, 37(11), 1134-1141. doi:10.1097/00004583-199811000-00012

Berger, Z., & Dalton, L. (2009). Coping with a cleft: psychosocial adjustment of adolescents with a cleft lip and palate and their parents. *Cleft Palate-Craniofacial Journal*, 46(4), 435-443. doi:10.1597/08-093.1

Bowen, M. (1978). *Family therapy in clinical practice*. London: Jason Aronson.

Bowlby J. (1969). *Attachment. Attachment and loss: Vol. 1. Loss*. New York: Basic Books.

Bradbury, E. T., & Hewison, J. (1994). Early parental adjustment to visible congenital disfigurement. *Child: Care, Health and Development*, 20(4), 251-266.

British Psychological Society (2007). *New ways of working for applied psychologists in health and social care: Working psychologically in teams*. Leicester: Author.

Bucci, S., Roberts, N. H., Danquah, A. N., & Berry, K. (2015). Using attachment theory to inform the design and delivery of mental health services: A systematic review of the literature. *Psychology and Psychotherapy: Theory, Research and Practice*, 88(1), 1-20. doi:10.1111/papt.12029

Byrnes, A., Berk, N., Cooper, M., & Marazita, M. (2003). Parental evaluation of informing interviews for cleft lip and/or palate. *Pediatrics*, 112(2), 308-313. Retrieved from <http://pediatrics.aappublications.org/content/112/2/308>

Centre for Reviews and Dissemination (2009). *Systematic Reviews: CRD's guidance for undertaking reviews in health care*. York: University of York. Retrieved from https://www.york.ac.uk/media/crd/Systematic_Reviews.pdf

Changing Faces. (2017). *Face equality*. Retrieved 10 January 2017, from <https://www.changingfaces.org.uk/resources/face-equality>

Cleft Lip and Palate Association. (2017). *Diagnosis and birth*. Retrieved from <https://www.clapa.com/treatment/diagnosis-birth/#antenatal>

Collett, B. R., Cloonan, Y. K., Speltz, M. L., Anderka, M., & Werler, M. M. (2012). Psychosocial functioning in children with and without orofacial clefts and their parents. *The Cleft Palate Craniofacial Journal*, 49, 397-405. doi:10.1597/10-007

Crawford, M. J., Rutter, D., Manley, C., Weaver, T., Bhui, K., Fulop, N., & Tyrer, P. (2002). Systematic review of involving patients in the planning and development of health care. *BMJ (Clinical Research Ed.)*, 325(7375), 1263. Retrieved from <http://www.bmj.com/>

- Department of Health. (2015). The NHS Constitution for England. Retrieved from https://www.gov.uk/government/uploads/system/uploads/attachment_data/file/480482/NHS_Constitution_WEB.pdf
- Division of Clinical Psychology (2011). *Good practice guidelines on the use of psychological formulation*. Leicester: British Psychological Society.
- Dixon-Woods, M., Booth, A., & Sutton, A. J. (2007). Synthesizing qualitative research: A review of published reports. *Qualitative Research*, 7(3), 375-422.
doi:10.1177/1468794107078517
- Drotar, D. (1997). Relating parent & family functioning to the psychological adjustment of children with chronic health conditions: What have we learned? What do we need to know? *Journal of Pediatric Psychology*, 22(2), 149-165. doi:10.1093/jpepsy/22.2.149
- Duggleby, W., Holtslander, L., Kylma, J., Duncan, V., Hammond, C., & Williams, A. (2010). Metasynthesis of the hope experience of family caregivers of persons with chronic illness. *Qualitative Health Research*, 20(2), 148-158.
doi:10.1177/1049732309358329
- Eiserman, W. (2001). Unique outcomes and positive contributions associated with facial difference: expanding research and practice. *Cleft Palate-Craniofacial Journal*, 38(3), 236-244. doi:10.1597/1545-1569(2001)038<0236:UOAPCA>2.0.CO;2
- Erikson, E. (1968). *Identity, youth and crisis*. New York: Norton.
- Fajardo, B. (1987). Parenting a damaged child: mourning, regression, and disappointment. *Psychoanalytic Review*, 74(1), 19-43.

- Feragen, K. B., & Stock, N. M. (2014). When there is more than a cleft: psychological adjustment when a cleft is associated with an additional condition. *The Cleft Palate-Craniofacial Journal*, *51*(1), 5-14. doi:10.1597/12-328
- Gassling, V., Christoph, C., Wahle, K., Koos, B., Wiltfang, J., Gerber, W.-D., & Siniatchkin, M. (2014). Children with a cleft lip and palate: An exploratory study of the role of the parent child interaction. *Journal of Cranio-Maxillo-Facial Surgery*, *42*, 963–968. doi:10.1016/j.jcms.2014.01.016
- Gilbert, P. (2009). Introducing Compassion Focused Therapy. *Advances in Psychiatric Treatment*, *15*, 199-208. doi:10.1192/apt.bp.107.005264
- Goldberg, S., Janus, M., Washington, J., Simmons, R. J., MacLusky, I., & Fowler, R. S. (1997). Prediction of preschool behavioral problems in healthy and pediatric samples. *Journal Of Developmental And Behavioral Pediatrics*, *18*(5), 304-313. doi:10.1097/00004703-199710000-00004
- Goodwin, I., Holmes, G., Cochrane, R., & Mason, O. (2003). The ability of adult mental health services to meet clients' attachment needs: the development and implementation of the Service Attachment Questionnaire. *Psychology And Psychotherapy*, *76*(Pt 2), 145-161. doi: 10.1348/147608303765951186
- *Hopkins, E. E., Gazza, E., & Marazita, M. L. (2016). Parental experience caring for cleft lip and palate infants with nasoalveolar moulding. *Journal of Advanced Nursing*, *72*(10), 2413-2422. doi:10.1111/jan.12994
- *Hsieh, Y., Chao, Y. Y., & Shiao, J. S. (2013). A qualitative study of psychosocial factors affecting expecting mothers who choose to continue a cleft lip and/or palate pregnancy to term. *The Journal of Nursing Research: JNR*, *21*(1), 1-9. doi:10.1097/jnr.0b013e3182828dbd

- Hunt, O., Burden, D., Hepper, P., & Johnston, C. (2005). The psychosocial effects of cleft lip and palate: a systematic review. *European Journal of Orthodontics*, *27*(3), 274-285. doi:10.1093/ejo/cji004
- Hunt, O., Burden, D., Hepper, P., Stevenson, M., & Johnston, C. (2007). Parent reports of the psychosocial functioning of children with cleft lip and/or palate. *The Cleft Palate-Craniofacial Journal*, *44*, 304–311. doi:10.1597/05-205
- *Johansson, B., & Ringsberg, K. (2004). Parents' experiences of having a child with cleft lip and palate. *Journal of Advanced Nursing*, *47*(2), 165-173. doi:10.1111/j.1365-2648.2004.03075x
- Klein, T. P., Pope, A., & Abbott, R. (2014). Social Interactions of School-Aged Children with CFA: Mothers' Perspectives and Advice. *The Cleft Palate-Craniofacial Journal*, *51*(3), 300-307. doi:10.1597/12-178
- Klein, T., Pope, A., Getahun, E., & Thompson, J. (2006). Mothers' reflections on raising a child with a craniofacial anomaly. *Cleft Palate-Craniofacial Journal*, *43*(5), 590-597. doi:10.1597/05-117
- *Lindberg, N., & Berglund, A. (2014). Mothers' experiences of feeding babies born with cleft lip and palate. *Scandinavian Journal of Caring Sciences*, *28*(1), 66-73. doi:10.1111/scs.12048
- Maris, C. L., Endriga, M. C., Speltz, M. L., Jones, K., & DeKlyen, M. (2000). Are infants with orofacial clefts at risk for insecure mother-child attachments? *The Cleft Palate-Craniofacial Journal*, *37*(3), 257-265. doi:10.1597/1545-1569(2000)037<0257:AIWOCA>2.3.CO;2

- Martin, V. (2005). Prenatal cleft lip and palate parent programme - phase I. *British Journal of Midwifery*, 13(2), 90-95. doi:10.12968/bjom.2005.13.2.17467
- *Martins, T. U., Zerbetto, S. R., & Dupas, G. (2013). Empowerment mechanisms used by the family of a child with a cleft lip and palate to a resilient path. *Ciencia, Cuidado E Saude*, 12(3), 494-501. doi:10.4025/ciencucuidsaude.v12i3.19577
- Mason, J. (2002). *Qualitative research*. London: Sage.
- Maxwell, J. A. (2012). *A realist approach for qualitative research*. London: Sage.
- *McCorkell, G., McCarron, C., Blair, S., & Coates, V. (2012). Parental experiences of cleft lip and palate services. *Community Practitioner*, 85(8), 24-27.
- Minuchin, S. (1974). *Families and family therapy*. Cambridge, MA: Harvard University Press.
- Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2010). Preferred reporting items for systematic reviews and meta-analyses: the PRISMA statement. *International Journal of Surgery*, 8(5), 336-341. doi:10.1016/j.ijssu.2010.02.007
- Murray, L., Hentges, F., Hill, J., Karpf, J., Mistry, B., Kreutz, M., ... Cleft Lip and Palate Study Team (2008). The effect of cleft lip and palate and the timing of lip repair on mother-infant interactions and infant development. *The Journal of Child Psychology and Psychiatry*, 49, 115–123. doi:10.1111/j.1469-7610.2007.01833x
- Nelson, J., O'Leary, C., & Weinman, J. (2009). Causal attributions in parents of babies with a cleft lip and/or palate and their association with psychological well-being. *The Cleft Palate-Craniofacial Journal*. 46(4), 425-434. doi:10.1597/07-194.1
- Nelson, P. (2009). Qualitative approaches in craniofacial research. *Cleft Palate-Craniofacial Journal*, 46(3), 245-251. doi:10.1597/08-121.1

- *Nelson, P. A., Caress, A., Glenny, A., & Kirk, S. A. (2012). 'Doing the "right" thing': how parents experience and manage decision-making for children's 'normalising' surgeries. *Social Science & Medicine*, 74(5), 796-804. doi:10.1016/j.socscimed.2011.11.024
- Nelson, P., Glenny, A., Kirk, S., & Caress, A. (2011). Parents' experiences of caring for a child with a cleft lip and/or palate: a review of the literature. *Child: Care, Health And Development*, 38(1), 6-20. doi:10.1111/j.1365-2214.2011.01244.x
- Nelson, P. A., & Kirk, S. A. (2013). Parents' Perspectives of Cleft Lip and/or Palate Services: A Qualitative Interview. *The Cleft Palate-Craniofacial Journal*, 50(3), 275-285. doi:10.1597/11-293
- *Nelson, P. A., Kirk, S. A., Caress, A., & Glenny, A. (2012). Parents' Emotional and Social Experiences of Caring for a Child Through Cleft Treatment. *Qualitative Health Research*, 22(3), 346-359. doi:10.1177/1049732311421178
- Noblit, G. W. & Hare, R. D. (1988). *Meta-Ethnography: Synthesizing qualitative studies*. London: Sage.
- *Nusbaum, R., Grubs, R., Losee, J., Weidman, C., Ford, M., & Marazita, M. (2008). A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period. *Journal of Genetic Counselling*, 17(4), 336-350. doi:10.1007/s10897-008-9152-5
- Phares, V., Lopez, E., Fields, S., Kamboukos, D., & Duhig, A. M. (2005). Are fathers involved in pediatric psychology research and treatment? *Journal of Pediatric Psychology*, 30(8), 631-643 doi:10.1093/jpepsy/jsi050

- Popay, J., Rogers, A., & Williams, G. (1998). Rationale and standards for the systematic review of qualitative literature in health services research. *Qualitative Health Research*, 8(3), 341-351. doi:10.1177/104973239800800305
- Pope, A., Tillman, K. & Snyder, H. T. (2005). Parenting stress in infancy and psychosocial adjustment in toddlerhood: a longitudinal study of children with craniofacial anomalies. *The Cleft Palate-Craniofacial Journal*, 42, 556–559. doi:10.1597/04-066R.1
- Public Health Resource Unit (2006). *Critical Appraisal Skills Programme(CASP): Making sense of evidence*. England: Public Health Resource Unit. Retrieved from http://www.casp.uk.net/wpcontent/uploads/2011/11/CASP_Qualitative_Appraisal_Checklist_14oct10.pdf
- Rey-Bellet, C., & Hohlfeld, J. (2004). Prenatal diagnosis of facial clefts: evaluation of a specialised counselling. *Swiss Medical Weekly*, 134(43-44), 640-644. Retrieved from <https://smw.ch/>
- Ring, N., Jepson, R., & Ritchie, K. (2011). Methods of synthesizing qualitative research studies for health technology assessment. *International Journal of Technology Assessment In Health Care*, 27(4), 384-390. doi:10.1017/S0266462311000389
- Rumsey, N., & Harcourt, D. (2004). Body image and disfigurement: Issues and interventions. *Body Image*, 1(1), 83-97. doi:10.1016/S1740-1445(03)00005-6
- Sandelowski, M., Docherty, S., & Emben, C. (1997). Qualitative metasynthesis: Issues and techniques. *Research in Nursing & Health*, 20(4), 365-371. doi:10.1002/(SICI)1098-240X(199708)20:4<365:AID-NUR9>3.0.CO;2-E

- Schreiber, R., Crooks, D., & Stern, P. (1997). Qualitative meta-analysis. In J. Morse (eds.), *Completing a qualitative project: Details and dialogue* (pp. 311–326). Thousand Oaks: Sage.
- SCOPE, (2003). *Right from the start template: Good practice in sharing the news*. London: Department of Health.
- Seager, M. (2014). Using attachment theory to inform psychologically minded care services, systems and environments. In A. N. Danquah & K. Berry (Eds.). *Attachment theory in adult mental health: A guide to clinical practice* (pp. 213-224). Oxon: Routledge
- Searle, A., Ryan, S., & Waylen, A. (2016). Health professional communication and the diagnosis and care of infants born with cleft lip and palate in the U.K. *Journal of Neonatal Nursing*, 22(5), 236-243. doi:10.1016/j.jnn.2016.04.005
- Sharif, M. O., Callery, P., & Tierney, S. (2013). The perspectives of children and young people living with cleft lip and palate: a review of qualitative literature. *The Cleft Palate-Craniofacial Journal*, 50(3), 297-304. doi:10.1597/12-054
- *Shipe, M. E., Edwards, T. C., Evans, K. N., Schook, C. C., Leavitt, D., Peter, A., & ... Tse, R. (2016). Optimizing Surgical Treatment of Internationally Adopted Children with Cleft Lip and/or Palate: Understanding the Family Experience. *The Cleft Palate-Craniofacial Journal*, 53(4), 444-452. doi:10.1597/14-260
- *Sischo, L., Broder, H. L., & Phillips, C. (2015). Coping with Cleft: A Conceptual Framework of Caregiver Responses to Nasoalveolar Molding. *The Cleft Palate-Craniofacial Journal*, 52(6), 640-650. doi:10.1597/14-113

- *Sischo, L., Clouston, S. P., Phillips, C., & Broder, H. L. (2016). Caregiver responses to early cleft palate care: A mixed method approach. *Health Psychology, 35*(5), 474-482. doi:10.1037/hea000026
- Slifer, K. J., Amari, A., Diver, T., Hilley, L., Beck, M., Kane, A., & McDonnell, S. (2004). Social interaction patterns of children and adolescents with and without oral clefts during a videotaped analogue social encounter. *The Cleft Palate-Craniofacial Journal 41*(2), 175-184. doi:10.1597/02-084
- Speltz, M. L., Arnsden, G. C., & Clarren, S. S. (1990). Effects of craniofacial birth defects on maternal functioning postinfancy. *Journal of Pediatric Psychology, 15*(2), 177-196. doi:10.1093/jpepsy/15.2.177
- Stock, N. M., & Feragen, K. B. (2016). Psychological adjustment to cleft lip and/or palate: A narrative review of the literature. *Psychology & Health, 31*(7), 777-813. doi:10.1080/08870446.2016.1143944
- Stock, N. M., Hammond, V., Owen, T., Kiff, J., Shanly, A., & Rumsey, N. (2016). Achieving Consensus in the Measurement of Psychological Adjustment to Cleft Lip and/or Palate. *The Cleft Palate-Craniofacial Journal, 53*(4), 421-426. doi:10.1597/15-071
- *Stock, N. M., & Rumsey, N. (2015). Parenting a child with a cleft: the father's perspective. *The Cleft Palate-Craniofacial Journal, 52*(1), 31-43. doi:10.1597/13-035
- *Stone, M. B., Botto, L. D., Feldkamp, M. L., Smith, K. R., Roling, L., Yamashiro, D., & Alder, S. C. (2010). Improving quality of life of children with oral clefts: perspectives of parents. *The Journal of Craniofacial Surgery, 21*(5), 1358-1364. doi:10.1097/SCS.0b013e3181ec6872

- Swedlund, M. P., Schumacher, J. B., Young, H. N., & Cox, E. D. (2012). Effect of communication style and physician–family relationships on satisfaction with pediatric chronic disease care. *Health Communication, 27*(5), 498-505. doi:10.1080/10410236.2011.616632
- Taib, B. G., Taib, A. G., Swift, A. C., & van Eeden, S. (2015). Cleft lip and palate: diagnosis and management. *British Journal of Hospital Medicine, 76*(10), 584. doi:10.12968/hmed.2015.76.10.584
- Tiemens, K., Nicholas, D., & Forrest, C. R. (2013). Living with difference: Experiences of adolescent girls with cleft lip and palate. *The Cleft Palate-Craniofacial Journal, 50*, 27–34. doi:10.1597/10-278
- *Tierney, S., Blackhurst, M., Scahill, R., & Callery, P. (2015). Loss and rebuilding: A qualitative study of late diagnosis of cleft palate. *Journal for Specialists in Pediatric Nursing, 20*(4), 280-289. doi:10.1111/jspn.12125
- 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 Medical Research Methodology, 14*80. doi:10.1186/1471-2288-14-80
- Trenchard, S., Taylor, C., Halpin, L., Williams, J., Phare D. & Douglas L. (2014. April). *Service-user evaluation of an antenatal group for parents expecting a baby with a cleft lip and/or palate*. Poster presented at the meeting of the Craniofacial Annual Scientific Conference, Oxford.
- *Vanz, A. P., & Ribeiro, N. R. (2011). Listening to the mothers of individuals with oral fissures. *Revista Da Escola De Enfermagem Da USP, 45*(3), 596-602. doi: 10.1590/S0080-62342011000300007

Walsh, D., & Downe, S. (2005). Meta-synthesis method for qualitative research: A literature review. *Journal Of Advanced Nursing*, 50(2), 204-211. doi:10.1111/j.1365-2648.2005.03380.x

Wardle, C., & Boyce, T. (2009). *Media coverage and audience reception of disfigurement on television*. London: The Healing Foundation and Cardiff University.

*Williams, L., Dures, E., Waylen, A., Ireland, T., Rumsey, N., & Sandy, J. (2012). Approaching parents to take part in a cleft gene bank: a qualitative pilot study. *The Cleft Palate-Craniofacial Journal*, 49(4), 425-436. doi:10.1597/10-086

World Health Organization. (2012). *Oral health (Fact Sheet No. 318)*. Retrieved from <http://www.who.int/mediacentre/factsheets/fs318/>

Yardley, L. (2008). Demonstrating validity in qualitative psychology. In J. A. Smith (Eds.), *Qualitative psychology: A practical guide to research methods* (pp. 235-251). London: Sage.

Young, J., O'Riordan, M., Goldstein, J., & Robin, N. (2001). What information do parents of newborns with cleft lip, palate, or both want to know? *Cleft Palate-Craniofacial Journal*, 38(1), 55-58. doi.org/10.1597/1545-1569(2001)038<0055:WIDPON>2.0

*Zeytinoğlu, S., Davey, M. P., Crerand, C., & Fisher, K. (2016). Fathers of children born with cleft lip and palate: Impact of the timing of diagnosis. *Families, Systems & Health: The Journal of Collaborative Family Healthcare*, 34(2), 150-158. doi:10.1037/fsh0000198

Figure 1: Flow diagram of the systematic search process following PRISM guidelines

(Moher, Liberati, Tetzlaff & Altman, 2010).

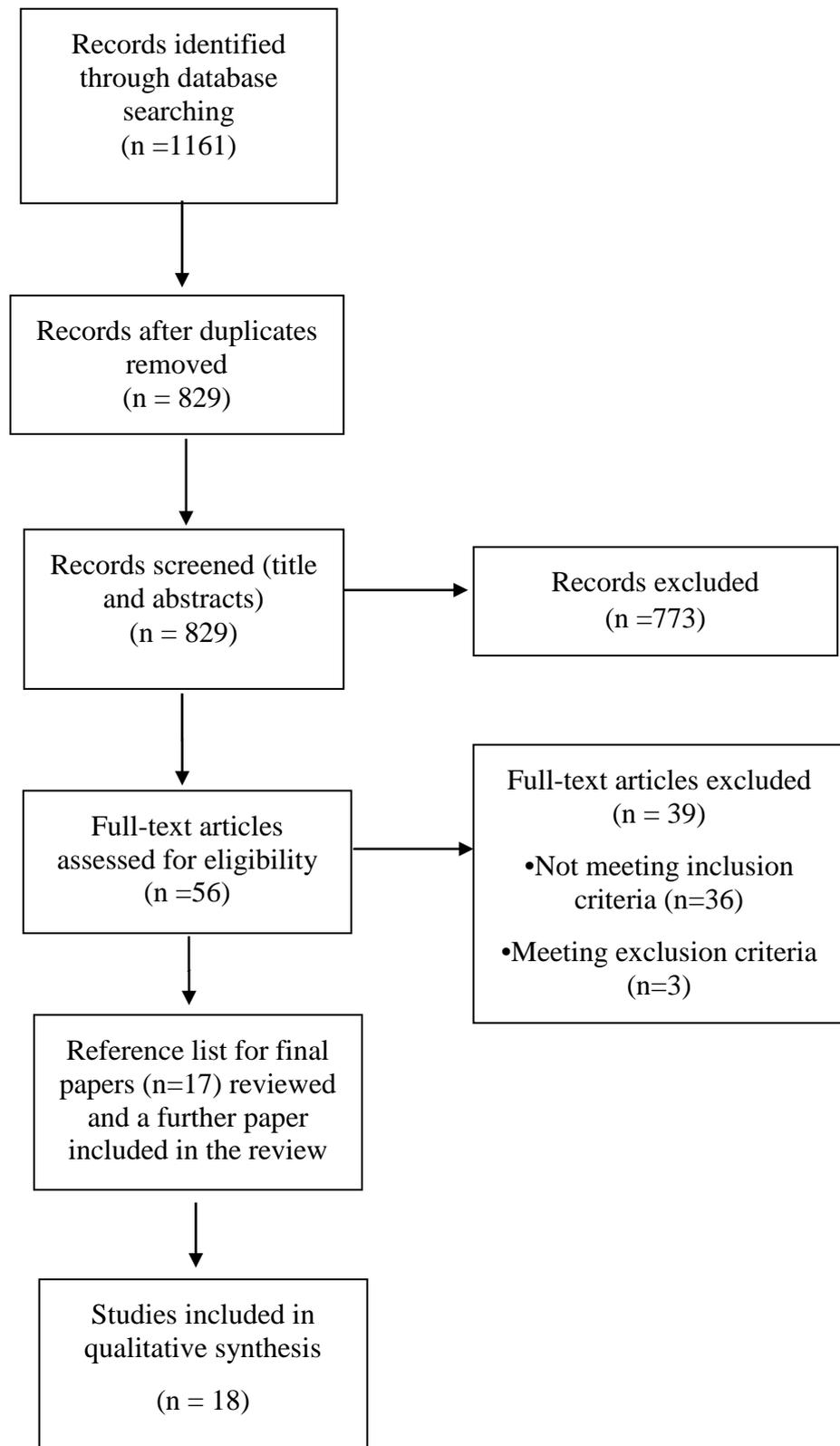


Table 1: *Summary information for the papers selected for synthesis*

Authors	Research Question/Aim	Methodology	Participants	Country of Origin
Hopkins, Gazza & Marazita (2016)	To explore the experience of parents caring for a child with a cleft lip and/or palate receiving nasosolevolar mounding.	Phenomenological	12 parents of children with a cleft lip and/or palate receiving nasosolevolar mounding, or had completed nasosolevolar mounding process in the past year	USA
Hsieh, Chao & Shiao (2013)	To identify the principal psychosocial factors that affect expecting mothers who choose to continue their cleft lip and/or palate pregnancy.	A qualitative phenomenographic approach	6 women who were expecting children who had been diagnosed with a cleft lip and/or palate	Taiwan
Johansson & Ringsberg (2004)	To describe parent's experiences of having a child with a cleft lip and/or palate.	A phenomenographic approach	20 families (20 mothers & 12 fathers) who had at least one child with a cleft lip and/or palate. Children aged between 1.5 months and 5 years old.	Sweden
Lindberg & Berglund (2014)	To describe the experiences of feeding for mothers of children born with a cleft lip and/or palate and to elucidate how they cope with challenges related to feeding.	Phenomenographic analysis	12 mothers of babies with different type of cleft diagnoses aged between 24 and 34 years' old. Their children were between the ages of 3 and 13 months old.	Norway
Martins, Zerbetto & Dupas (2013)	To know the empowerment mechanisms that families of children with a cleft lip and palate have developed or enhanced to be resilient when facing difficulties.	Content analysis	10 families of children with a cleft lip and palate of up to 3 years of age. 5 interviews were conducted with both the mother and father, and 5 interviews with just the mother.	Brazil
McCorkell, McCarron, Blair & Coates (2012)	To clarify the information and support most valued by parents in receipt of support from the cleft liaison nurse service and to identify service improvements.	Thematic analysis	16 mothers, 1 father and 1 grandmother of children with a cleft lip and/or palate. Children's ages ranged from 12 months to 8 years of age.	United Kingdom
Nelson, Caress, Glennly & Kirk (2012)	To explore how mothers and fathers experience and manage decision making during their child's cleft treatment.	Grounded theory	27 families (35 parents) with a child who a cleft lip and/or palate. Child's age ranged from 20 weeks to 21 years old. 8 couples interviewed together, the rest separately.	United Kingdom
Nelson, Kirk, Caress & Glennly (2012)	To explore the emotional and social experiences of mothers and fathers caring for a child with a cleft lip and/or palate.	Grounded theory	27 families (35 parents) with a child who a cleft lip and/or palate. Child's age ranged from 20 weeks to 21 years old. 8 couples interviewed together, the rest separately.	United Kingdom

Nusbaum et al. (2008)	To investigate the experience of receiving a diagnosis of cleft lip and/or palate in the prenatal or postnatal period	Qualitative descriptive approach with an emphasis on thematic analysis	20 parents of children with a cleft lip and/or palate. 12 parents received a prenatal diagnosis (including 4 mother-father couples), and 8 parents who received their child's diagnosis postnatal (including 3 mother-father couples)	USA
Shipe et al. (2016)	To understand the experience of families with children undergoing cleft surgery following adoption from a country outside the United States.	Mixed method design. A grounded theory approach used on the qualitative data.	20 parents of children with a cleft lip and/or palate who had been adopted outside the United States	USA
Sischo, Broder & Phillips (2015)	To present a conceptual framework of caregiver coping and adaptation to early cleft care using nasoalveolar moulding.	Modified grounded theory.	68 parents of children with cleft lip and/or palate. Parents interviewed at 3 timepoints. Child aged between 1 month and 13 months of age.	USA
Sischo, Clouston, Phillips & Broder (2016)	To understand caregiver's responses to early cleft lip/palate care for their child.	Mixed method design. Qualitative analysis using grounded theory	118 parents of children with a cleft lip and/or palate interviewed at 3 time points: the beginning of treatment; pre-surgery; post-surgery. Child between 1 and 13 months old.	USA
Stock & Rumsey (2015)	To explore the impact of having a child born with a cleft lip and/or palate from the father's perspective.	Thematic analysis	15 fathers aged between 31 and 58 years old. Majority of fathers had one child born with a cleft lip and/or palate, one father had two. Children's ages ranged from 4 to 24 years old.	United Kingdom
Stone et al. (2010)	To explore how parents' perceive the quality of life of their children with oral clefts and beliefs regarding factors affecting quality of life.	Unspecified thematic analysis	2 focus groups. One group consisted of 12 parents representing 12 children with a cleft lip and/or palate. One group consisted of 8 parents representing 5 children with a cleft lip and/or palate.	USA
Tierney, Blackhurst, Scahill & Callery (2015)	To explore the experiences of parents whose children received a late diagnosis of cleft palate.	Framework analysis	Parents of 17 children with a cleft lip and/or palate aged 4 year or younger. All children diagnosed with a cleft lip and/or palate more than 24 hours after being born. 3 nurses interviewed	United Kingdom
Vanz & Ribeiro (2011)	To report on what mothers of children with cleft lip and/or palate know about its etiology and identify their beliefs.	Content analysis	8 mothers, aged between 18 and 45, of children diagnosed with isolated cleft lip and/or palate. Children aged between 1 and 5 years old.	Brazil

Williams et al. (2012)	To identify factors that may contribute to participation in a cleft gene bank and the issues that should be considered when approaching parents.	Thematic analysis	16 parents with children cleft lip and/or palate. 5 focus groups were held.	United Kingdom
Zeytinoglu, Davey, Crerand & Fisher (2016)	To examine how the timing of a child's diagnosis affects how father's cope and adapt to having a child with a cleft lip and/or palate.	Thematic analysis	17 fathers (10 experienced a prenatal diagnosis, 7 a postnatal diagnosis) of children with a cleft lip and/or palate. Child aged between 1 and 4 years old.	USA

Text in the tables was taken directly from the reviewed papers

Appendix A: Search Strategy

Database Search Terms

	Child's Diagnosed Condition	Study Population	Type of Data Collected	Data Collection and Analysis
Search terms (combined using Boolean operator "OR" within terms and "AND" across terms)	<i>MEDLINE</i> ("Cleft Lip" or "Cleft Palate") <i>PsychINFO</i> ("Cleft Palate") <i>CINAHL</i> ("Cleft Lip") OR ("Cleft Palate") <i>AMED</i> ("Cleft lip") OR ("Cleft palate")	<i>MEDLINE</i> ("Parents+") <i>PsychINFO</i> ("Parents+") <i>CINAHL</i> ("Parents+") <i>AMED</i> ("Parents") OR ("Parent Child Relations") OR ("Parenting")	<i>MEDLINE</i> ("Life Change Events") <i>PsychINFO</i> ("Life Experiences") <i>CINAHL</i> ("Life Experiences")	<i>MEDLINE</i> ("Qualitative Research") <i>PsychINFO</i> ("Qualitative Research") <i>CINAHL</i> ("Qualitative Studies")
	Free text search terms cleft lip and palate cleft lip cleft palate orofacial cleft* craniofacial cranio-facial clefting CLP	Free text search terms parent* mother* father* caregiver famil* carer guardian child* infant* adolecen* young person*	Free text search terms experience* life experiences life change* needs perceptions stories views concern voice perspective feel understand worr*	Free text search terms Findings qualitative interview* grounded theory phenomenolog* narrative thematic analysis discourse semi-structured indepth focus groups discussion

+ = search term exploded

Appendix B: CASP Scores

Study	Research design	Sampling	Data collection	Reflexivity	Ethical issues	Data analysis	Findings	Value of research	Overall
Hopkins et al. (2016)	3	3	3	2	3	3	3	3	23
Hsieh et al. (2013)	1	2	3	2	3	2	3	3	19
Johansson & Ringsberg (2004)	2	3	2	1	3	2	3	3	19
Lindberg & Berglund (2014)	2	2	3	1	2	3	3	3	19
Martins et al. (2013)	3	2	3	2	2	2	3	2	19
McCorkell et al. (2012)	1	3	3	1	1	1	3	2	15
Nelson, Caress, et al. (2012)	3	3	2	1	1	2	3	3	18
Nelson, Kirk, et al. (2012)	3	3	3	2	3	3	3	3	23
Nusbaum et al. (2008)	3	2	3	2	3	3	3	3	22
Shipe et al. (2016)	3	3	3	1	2	3	3	3	21
Sischo et al. (2015)	3	3	3	1	2	3	3	3	21
Sischo et al. (2016)	3	3	3	1	1	3	3	3	20
Stock & Rumsey (2015)	3	3	3	1	3	3	3	3	22

Stone et al. (2010)	3	3	3	1	1	1	2	3	17
Tierney et al. (2015)	3	3	3	2	2	3	3	3	22
Vanz & Ribeiro (2011)	1	2	2	1	2	1	2	2	13
Williams et al. (2012)	3	3	3	3	3	3	3	3	24
Zeytinoglu et al. (2016)	2	3	3	1	2	3	3	3	20

1 = little or no justification or explanation for a particular area.

2 = moderate justification or explanation but not fully elaborated on.

3 = strong justification or explanation and explained relevant issue at hand.

Appendix C: Theme Extraction

Authors	Theme Extraction
Hopkins et al. (2016)	<ol style="list-style-type: none"> 1. You do what you have to do 2. We weren't left alone in the dark
Hsieh et al. (2013)	<ol style="list-style-type: none"> 1. Loss of self-value as a mother 2. Blaming the mother 3. Indecision about continuing pregnancy 4. Anxiety triggered by insufficient information 5. Burden of care and concerns about potential disadvantage status
Johansson & Ringsberg (2004)	<ol style="list-style-type: none"> 1. Unexpected event having a child with CLP 2. Reactions
Lindberg & Berglund (2014)	<ol style="list-style-type: none"> 1. Being a capable and good mother 2. Coping with the challenges related to feeding
Martins et al. (2013)	<ol style="list-style-type: none"> 1. Living one day at a time: winning steps 2. Supporting and being supported
McCorkell et al. (2012)	<ol style="list-style-type: none"> 1. Emotional experiences of having a child with a cleft
Nelson, Caress, et al. (2012)	<ol style="list-style-type: none"> 1. Core category: Doing the "right" thing: the moral dimension in decisions for children's surgery Subcategories: <ul style="list-style-type: none"> • 'Doing something': pursuing solutions • 'Fitting in': surgical normalisation • 'Being in the "right" hands: parents trust in cleft care practitioners
Nelson, Kirk, et al. (2012)	<ol style="list-style-type: none"> 1. Core category: Doing the "right" thing: the moral dimension in decisions for children's surgery Subcategory "managing emotions" <ul style="list-style-type: none"> • Conflicting emotions • The uncertainty of long-term treatment • Stigmatizing reactions to a child's impairment
Nusbaum et al. (2008)	<ol style="list-style-type: none"> 1. Receiving the diagnosis 2. Cleft Cause and embodied knowledge 3. Parent to parent support 4. Disability 5. Coping 6. Religion as an aid in coping 7. Preparation 8. Disadvantages of a prenatal diagnosis 9. Alternative perspectives
Shipe et al. (2016)	<ol style="list-style-type: none"> 1. Parental anxieties surrounding surgery 2. Considerations of the timing of surgery 3. Impact of the surgical experience
Sischo et al. (2015)	<ol style="list-style-type: none"> 1. NAM and the process of empowerment <ul style="list-style-type: none"> • Initial anxiety and self-doubt • Self-efficacy and NAM • NAM and positive identity construction • Role of social support
Sischo et al. (2016)	<ol style="list-style-type: none"> 1. NAM treatment 2. Lip surgery

- | | |
|--------------------------|---|
| | <ol style="list-style-type: none">3. Capabilities/coping4. Family environment/adaptation |
| Stock & Rumsey (2015) | <ol style="list-style-type: none">1. Variations in care and support2. Appraisals of the cleft3. Perceptions of treatment4. Looking back and moving forward |
| Stone et al. (2010) | <ol style="list-style-type: none">1. Birth and diagnosis2. Seeking information3. Surgery4. Support5. Psychosocial issues6. Finances7. Emotional impact on parents |
| Tierney et al. (2015) | Loss and Rebuilding: <ol style="list-style-type: none">1. Crescendo2. Bombshell3. Aftermath |
| Vanz & Ribeiro (2011) | <ol style="list-style-type: none">1. Mothers reaction2. Mothers feeling3. Knowledge of cause4. Teams behaviour |
| Williams et al. (2012) | <ol style="list-style-type: none">1. New knowledge has to be a good thing2. The pressures of early days3. Relationships with experts |
| Zeytinoglu et al. (2016) | <ol style="list-style-type: none">1. First hearing the diagnosis2. Taking care of a baby with cleft lip or palate3. Future concerns4. Reflections |
-

Appendix D: Table of Themes and Key Concepts

Study	A New Idea of Parenthood: Working Through the Emotional Experience	Becoming Immersed Within a New and Unexpected Medical System	Treatment Pathways and the Developing Parent Identity	Anticipating and Managing the Reactions of Others	Meeting Parenting Challenges Through Personal Growth and Support
Hopkins et al. (2016)		We Weren't Left Alone in the Dark	You Do What You Have To Do		We Weren't Left Alone in the Dark
Hsieh et al. (2013)	Loss of Self Value as a Mother; Blaming the Mother	Anxiety Triggered by Insufficient Information	Indecision About Continuing Pregnancy; Burden of Care and Concerns about Potential Disadvantage Status	Indecision About Continuing Pregnancy	
Johansson & Ringsberg (2004)	Unexpected Event of Having a Child with Cleft Lip and/or Palate	Unexpected Event of Having a Child with Cleft Lip and/or Palate	Reactions	Reactions	
Lindberg & Berglund (2014)	Being a Capable and Good Mother; Coping with Challenges Related to Feeding	Coping with Challenges Related to Feeding	Being a Capable and Good Mother		Coping with Challenges Related to Feeding
Martins et al. (2013)	Living One Day at a Time: Winning Steps	Living One Day at a Time: Winning Steps			Living One Day at a Time: Winning Steps; Supporting and Being Supported

McCorkell et al. (2012)	Emotional Experiences of Having a Child with a Cleft	Emotional Experiences of Having a Child with a Cleft			
Nelson, Caress, et al. (2012)		Being in the “right” hands: Parents trust in Cleft Care Practitioners	‘Doing Something’: Pursuing Solutions; ‘Fitting in’; Surgical Normalisation; Being in the “right” hands: Parents Trust in Cleft Care Practitioners	‘Doing Something’: Pursuing Solutions; ‘Fitting in’; Surgical Normalisation	
Nelson, Kirk, et al. (2012)	Conflicting Emotions	Conflicting Emotions; The Uncertainty of Long Term Treatment.	The Uncertainty of Long Term Treatment.	Stigmatizing Reactions to a Childs Impairment	
Nusbaum et al. (2008)	Cleft Cause and Embodied Knowledge; Coping; Disadvantages of a Prenatal Diagnosis; Alternative Perspectives	Receiving the Diagnosis; Preparation		Disability	Parent-to-Parent Support; Religion as an Aid in Coping
Shipe et al. (2016)		Parental Anxieties Surrounding Surgery	Considerations of the Timing of Surgery; Impact of the Surgical Experience		
Sischo et al. (2015)		NAM and Process of Empowerment: Initial Anxiety and Self Doubt; Role of	NAM and Process of Empowerment: Self Efficacy and NAM; NAM and Positive		NAM and Process of Empowerment: Role of Social Support

		Social Support	Identity Construction		
Sischo et al. (2016)		Capabilities/Coping; NAM Treatment	Lip Surgery	Family Environment/Adaptation; Capabilities/ Coping	Capabilities/Coping
Stock & Rumsey (2015)	Appraisals of the Cleft; Looking Back and Moving Forward	Variations in Care and Support; Perceptions of Treatment		Variations in Care and Support; Looking Back and Moving Forward	Looking Back and Moving Forward
Stone et al. (2010)	Birth and Diagnosis; Emotional Impact on Parents	Surgery; Finances; Seeking Information		Emotional Impact on Parents; Psychosocial Issues	Support
Tierney et al. (2015)	Crescendo; Bombshell	Crescendo; Aftermath			
Vanz & Ribeiro (2011)	Mothers Feeling; Mothers Reaction; Knowledge of the Cause	Teams Behaviour		Mothers Feeling	
Williams et al. (2012)	The Pressures of the Early Days	The Pressures of the Early Days; Relationships with Experts			New Knowledge Has to be a Good Thing
Zeytinoğlu et al. (2016)	First Hearing the Diagnosis; Taking Care of a Baby with a Cleft Lip or Palate		Taking Care of a Baby with a Cleft Lip or Palate	First Hearing the Diagnosis; Future Concerns	Reflections

Appendix E: Interpretative Analysis Audit**A New Idea of Parenthood: Working Through the Emotional Experience**

Extracted Themes and Key Concepts	Emerging Third-Order Concept	Final Third-Order Concept
Birth and Diagnosis; Mothers Feeling; Emotional Experiences of Having a Child with a Cleft; Unexpected Event of Having a Child with Cleft Lip and/or Palate; Coping; First Hearing the Diagnosis; Emotional Impact on Parents; Alternative Perspectives; The Pressures of the Early Days	A range of powerful emotions when child is diagnosed with the condition and when child is born	A New Idea of Parenthood: Working Through the Emotional Experience
Taking Care of a Baby with Cleft Lip or Palate; Appraisals of the Cleft; Cleft Cause and Embodied Knowledge; Crescendo; Bombshell; Living One Day at a Time: Winning Steps; Coping with Challenges Related to Feeding; Being a Capable and Good Mother	Concerns about the perceived challenges of the condition and parents' ability to manage	
Blaming the Mother; Knowledge of the Cause; Disadvantages of a Prenatal Diagnosis; Loss of Self-Value as a Mother; Birth and Diagnosis; Mothers Reaction; Coping; Looking Back and Moving Forward; Conflicting Emotions	Loss of the image of 'perfect' child and mother and period of adjustment	

Becoming Immersed Within a New and Unexpected Medical System

Extracted Themes and Key Concepts	Emerging Third-Order Concept	Final Third-Order Concept
<p>Teams Behaviour; Variations in Care and Support; Receiving the Diagnosis; Coping with Challenges Related to Feeding; Seeking Information; Preparation; Anxiety Triggered by Insufficient Information; Crescendo; Aftermath; ‘Being in the “right” hands’: Parents Trust in Cleft Care Practitioners; Capabilities/Coping; We Weren’t Left Alone in the Dark; NAM and Process of Empowerment: Role of Social Support; Relationships with Experts; Emotional Experiences of Having a Child with a Cleft; Unexpected Event of Having a Child with Cleft Lip and/or Palate</p>	<p>The impact of healthcare support and the healthcare system in containing parents’ emotions and influencing their perceived ability to cope. The role of perceived professional knowledge in reducing the feelings of disquiet</p>	<p>Becoming Immersed Within a New and Unexpected Medical System</p>
<p>The Pressures of the Early Days; Living One Day at a Time: Winning Steps; Parental Anxieties Surrounding Surgery; NAM Treatment; Surgery; Perceptions of Treatment; Finances; NAM and Process of Empowerment: Initial Anxiety and Self Doubt; Conflicting Emotions; The Uncertainty of Long Term Treatment</p>	<p>Pressures of treatment journey, can be experienced as emotionally overwhelming</p>	

Treatment Pathways and the Developing Parent Identity

Extracted Themes and Key Concepts	Emerging Third-Order Concept	Final Third-Order Concept
You Do What You Have To Do; Considerations of the Timing of Surgery; Impact of the Surgical Experience; Reactions; Indecision About Continuing Pregnancy; ‘Doing Something’: Pursuing Solutions; NAM and Process of Empowerment: Self Efficacy and NAM; Being a Capable and Good Mother; The Uncertainty of Long Term Treatment; Burden of Care and Concerns about Potential Disadvantage Status	Parents desire to do their best for their child, seemingly moral aspect to be the best parent they can be	Treatment Pathways and the Developing Parent Identity
Lip Surgery; ‘Fitting in’; Surgical Normalisation; Taking Care of a Baby with a Cleft Lip or Palate; ‘Being in the “right” hands’: Parents Trust in Cleft Care Practitioners; Considerations of the Timing of Surgery; NAM and Process of Empowerment: Positive Identity Construction	Parents engagement with treatment and healthcare professionals providing reassurance they are virtuous and responsible parents	

Anticipating and Managing the Reactions of Others

Extracted Themes and Key Concepts	Emerging Third-Order Concept	Final Third-Order Concept
Reactions; Family Environment/Adaptation Emotional Impact on Parents, Indecision About Continuing Pregnancy; Variations in Care and Support; Mothers Feeling.	Parents' experiences of both positive and negative reactions from friends, family and society	Anticipating and Managing the Reactions of Others
Stigmatizing Reactions to a Child's Impairment; Capabilities/Coping; Psychosocial Issues; Variations in Care and Support; First Hearing the Diagnosis; Looking Back and Moving Forward; 'Doing Something': Pursuing Solutions; 'Fitting in'; Surgical Normalisation; Future Concerns; Disability	Parental experiences leading to personal reflections around their own understandings of 'difference'. Wider societal views impacting future anticipatory concerns	

Meeting Parenting Challenges Through Personal Growth and Support

Extracted Themes and Key Concepts	Emerging Third-Order Concept	Final Third-Order Concept
Capabilities/Coping; Looking Back and Moving Forward; Reflections, Religion as an Aid in Coping; NAM and Process of Empowerment: Role of Social Support; Living One Day at a Time: Winning Steps; Coping with Challenges Related to Feeding	Personal growth reported, learning effective coping strategies	Meeting Parenting Challenges Through Personal Growth and Support
Supporting and Being Supported; Support; NAM and Process of Empowerment: Role of Social Support; We Weren't Left Alone in the Dark; Parent-to-Parent Support; New Knowledge Has to Be a Good Thing; Looking Back and Moving Forward	Appreciating the value of parent-to-parent support. Receiving and providing support	

Appendix F: Author Guidelines *Child: Care, Health and Development*

GENERAL

Child: Care, Health and Development is an international, peer-reviewed journal which publishes papers dealing with all aspects of the health and development of children and young people. We aim to attract quantitative and qualitative research papers relevant to people from all disciplines working in child health. We welcome studies which examine the effects of social and environmental factors on health and development as well as those dealing with clinical issues, the organization of services and health policy. We particularly encourage the submission of studies related to those who are disadvantaged by physical, developmental, emotional and social problems. The journal also aims to collate important research findings and to provide a forum for discussion of global child health issues.

Please read the instructions below carefully for details on the submission of manuscripts, the journal's requirements and standards as well as information concerning the procedure after a manuscript has been accepted for publication in *Child: Care, Health and Development*. Authors are encouraged to visit Wiley-Blackwell Author Services for further information on the preparation and submission of articles and figures.

3. MANUSCRIPT SUBMISSION PROCEDURE

Manuscripts should be submitted electronically via the online submission site <http://mc.manuscriptcentral.com/cch>. The use of an online submission and peer review site enables immediate distribution of manuscripts and consequentially speeds up the review process. It also allows authors to track the status of their own manuscripts. Complete instructions for submitting a paper is available online and below. Further assistance can be obtained from Editorial Assistant Iris Poesse at cchadmin@wiley.com.

A covering letter must be submitted as part of the online submission process, stating on behalf of all the authors that the work has not been published and is not being considered for publication elsewhere.

Important note: All papers will go through a initial sifting process within the editorial board.

3.1. Getting Started

- Launch your web browser (supported browsers include Internet Explorer 6 or higher, Netscape 7.0, 7.1, or 7.2, Safari 1.2.4, or Firefox 1.0.4) and go to the journal's online Submission Site: <http://mc.manuscriptcentral.com/cch>
- Log-in or click the 'Create Account' option if you are a first-time user.
- If you are creating a new account.
 - After clicking on 'Create Account', enter your name and e-mail information and click 'Next'. Your e-mail information is very important.
 - Enter your institution and address information as appropriate, and then click 'Next.'
 - Enter a user ID and password of your choice (we recommend using your e-mail address as your user ID), and then select your area of expertise. Click 'Finish'.
- If you have an account, but have forgotten your log in details, go to Password Help on the

journals online submission system <http://mc.manuscriptcentral.com/cch> and enter your e-mail address. The system will send you an automatic user ID and a new temporary password.

- Log-in and select 'Author Centre.'

3.2. Submitting Your Manuscript

- After you have logged in, click the submission link in the menu bar.
- Enter data and answer questions as appropriate. You may copy and paste directly from your manuscript and you may upload your pre-prepared covering letter.
- Click the 'Next' button on each screen to save your work and advance to the next screen.
- You are required to upload your files.
 - Click on the 'Browse' button and locate the file on your computer.
 - Select the designation of each file in the drop-down menu next to the 'Browse' button.
 - When you have selected all files you wish to upload, click the 'Upload Files' button.
- Review your submission (in HTML and PDF format) before sending it to the Journal. Click the 'Submit' button when you are finished reviewing.

3.3. Manuscript Files Accepted

Manuscripts should be uploaded as Word (.doc) or Rich Text Format (.rft) files (**not** write-protected) plus separate figure files. It is recommended that, where possible, line figures be embedded into a single Microsoft Word document. For halftone figures, only high-resolution TIF or EPS files are suitable for printing. The text file must contain the entire manuscript including Abstract (structured abstracts, not more than 300 words, including background, methods, results and conclusions are preferred); Introduction; Methods; Results; Discussion; Acknowledgements; References; Tables; Figure legends, but no embedded figures. Manuscripts should be formatted as described in the Author Guidelines below.

3.4. Peer Review Process

Manuscripts submitted to *Child: Care, Health and Development* are subject to initial scrutiny by the SIFT committee which consists of members of the Editorial Board. Where the SIFT Committee believe it unlikely that the paper will be acceptable for publication either for methodological reasons or because it does not fall within areas likely to be of central interest to our readers the paper will not be sent for formal peer review. The authors will be notified of this decision.

Manuscripts passing this initial scrutiny are reviewed by experts in the field, using a system of double-blinded review. The names of the reviewers will thus not be disclosed to the author submitting a paper and the name(s) of the author(s) will not be disclosed to the reviewers.

To allow double blinded review, please submit (upload) your main manuscript and title page as separate files.

Please upload:

- Your manuscript without title page under the file designation 'main document'
- Figure files under the file designation 'figures'
- The title page, Acknowledgements and Conflict of Interest Statement where applicable, should be uploaded under the file designation 'title page'

All documents uploaded under the file designation 'title page' will not be viewable in the HTML and PDF format you are asked to review in the end of the submission process. The files viewable in the HTML and PDF format are the files available to the reviewer in the review process.

3.5. Suggest a Reviewer

Child: Care, Health and Development attempts to keep the review process as short as possible to enable rapid publication of new scientific data. In order to facilitate this process, please suggest the names and current email addresses of 2 potential international reviewers whom you consider capable of reviewing your manuscript. In addition to your choice the journal editor will choose one or two reviewers as well.

3.6. Suspension of Submission Mid-way in the Submission Process

You may suspend a submission at any phase before clicking the 'Submit' button and save it to submit later. The manuscript can then be located under 'Unsubmitted Manuscripts' and you can click on 'Continue Submission' to continue your submission when you choose to.

3.7. E-mail Confirmation of Submission

After submission you will receive an e-mail to confirm receipt of your manuscript. If you do not receive the confirmation e-mail after 24 hours, please check your e-mail address carefully in the system. If the e-mail address is correct please contact your IT department. The error may be caused by some sort of spam filtering on your e-mail server. Also, the e-mails should be received if the IT department adds our e-mail server (uranus.scholarone.com) to their whitelist.

3.8. Manuscript Status

You can access ScholarOne Manuscripts (formerly known as Manuscript Central) any time to check your 'Author Centre' for the status of your manuscript. The Journal will inform you by e-mail once a decision has been made.

3.9. Submission of Revised Manuscripts

Revised manuscripts must be uploaded within 3 months of authors being notified of the decision. In exceptional cases a longer period may be agreed with the editor. Locate your manuscript under 'Manuscripts with Decisions' and click on 'Submit a Revision' to submit your revised manuscript. Please remember to delete any old files uploaded when you upload your revised manuscript. Please also remember to upload your manuscript document separate from your title page.

4. MANUSCRIPT TYPES ACCEPTED

Original Articles: Articles reporting original scientific data based quantitative or qualitative research are particularly welcomed. Articles should begin with a structured abstract and should ideally be between 2,000 and 3,000 words in length excluding tables and references. In the case of complex qualitative research reports, the editors may be prepared to extend the word limit to 5000 words.

Review Papers: The journal welcomes syntheses of research in the form of systematic

reviews. The word limit may be extended, in some circumstances, to 5000 words. Reviews are structured in the same way as original research (see above). The journal will occasionally publish narrative reviews where it is felt that these will be of particular interest to the readers and will be important in encouraging debate.

Case Reports: The journal will very occasionally publish case reports but only where these are believed by the editors to hold important generalisable lessons for the clinical or scientific community. We would expect such reports to begin with a very brief narrative abstract. The main text (1500 words maximum) should include a brief description of the case followed by a short discussion section explaining the implications of the case for clinical practice or research. Normal processes of peer review apply.

Short Communications: The journal will occasionally publish short communications. Typically these will report the results of relatively simple studies with straightforward analyses and results. The format may be flexible in discussion with the editors but will normally consist of an extremely brief abstract followed by a main text containing not more than 1500 words and not more than 2 tables or illustrations. Normal processes of peer review apply.

Letters to the Editor: We encourage letters to the editor, either in response to published articles or where authors wish to raise important areas for discussion amongst the readership. The decisions on whether or not to publish will normally be taken within the editorial board and are based on whether it is felt that the letter opens or continues an important area for scientific debate.

Editorials: From time to time the editors will commission editorials, often to accompany specific papers or groups of papers. The format for these editorials is individually negotiated. Authors may choose to submit an editorial in the form of a brief (1200 words maximum) discussion with not more than 15 references on any subject.

All submissions, including those commissioned by the editors are subject to external peer review.

Special Issues: From time to time the Editor will commission a special issue of the Journal which will take the form of a number of papers devoted to a particular theme.

5. MANUSCRIPT FORMAT AND STRUCTURE

5.1. Format

Units and spellings: Système International (SI) units should be used, as given in *Units, Symbols and Abbreviations* (4th edition, 1988), published by the Royal Society of Medicine Services Ltd, 1 Wimpole Street, London W1M 8AE, UK. Spelling should conform to that used in *The Concise Oxford Dictionary*, published by Oxford University Press.

Language: The language of publication is English. If English is not your first language, then you will find it helpful to enlist the help of a native English speaker to edit the piece, to correct grammar and ensure that idioms are correct. This too makes it easier for the reviewers to give full justice to your work. Authors for whom English is a second language may choose

to have their manuscript professionally edited before submission to improve the English. A list of independent suppliers of editing services can be found at http://authorservices.wiley.com/bauthor/english_language.asp. All services are paid for and arranged by the author, and use of one of these services does not guarantee acceptance or preference for publication.

Chinese Scholars Network is a resource for scholars, academics, and researchers in China who would like to publish their work in English language journals.

5.2. Structure

The following checklist should be used to check the manuscript before submission. Articles are accepted for publication at the discretion of the Editor. A manuscript reporting original research should ideally be between 2000 and 3000 words. In the case of complex qualitative research reports, or systematic reviews, the editors may in some circumstances be prepared to extend the word limit to 5000 words. The manuscript should consist of the sections listed below.

Title Page: The title page should give both a descriptive title and short title. The title should be concise and give a brief indication of what is in the paper. Authors are required to detail in full: qualifications, current job title, institution and full contact details. Also a word count for the article and keywords should be given on the title page.

To allow double-blinded review, please submit (upload) your main manuscript and title page as separate files as explained in section 3.4.

Abstract: Structured abstracts, not more than 300 words, including background, methods, results and conclusions are preferred

Optimizing Your Abstract for Search Engines

Many students and researchers looking for information online will use search engines such as Google, Yahoo or similar. By optimizing your article for search engines, you will increase the chance of someone finding it. This in turn will make it more likely to be viewed and/or cited in another work. We have compiled these guidelines to enable you to maximize the web-friendliness of the most public part of your article.

Main Text

Generally, all papers should be divided into the following sections and appear in this order: Abstract (structured abstracts, not more than 300 words, including background, methods, results and conclusions are preferred); Introduction; Methods; Results; Discussion; Acknowledgements (these should be brief and must include references to sources of financial and logistical support); References; Tables; Figures.

Key Messages

From 2007 onwards a key messages box should be provided with each manuscript. This should include up to 5 messages on key points of practice, policy or research. This also applies to articles solicited for themed issues.

5.3. References

References should be prepared according to **the *Publication Manual of the American***

Psychological Association (6th edition). This means in text citations should follow the author-date method whereby the author's last name and the year of publication for the source should appear in the text, for example, (Jones, 1998). The complete reference list should appear alphabetically by name at the end of the paper.

A sample of the most common entries in reference lists appears below. Please note that a DOI should be provided for all references where available. For more information about APA referencing style, please refer to the [APA FAQ](#). Please note that for journal articles, issue numbers are not included unless each issue in the volume begins with page one.

Journal article

Beers, S. R. , & De Bellis, M. D. (2002). Neuropsychological function in children with maltreatment-related posttraumatic stress disorder. *The American Journal of Psychiatry*, **159**, 483–486. doi:[10.1176/appi.ajp.159.3.483](https://doi.org/10.1176/appi.ajp.159.3.483)

Book edition

Bradley-Johnson, S. (1994). Psychoeducational assessment of students who are visually impaired or blind: Infancy through high school (2nd ed.). Austin, TX: Pro-ed.

Work that has not been accepted for publication and personal communications should not appear in the reference list, but may be referred to in the text (e.g. 'A. Author, unpubl. observ.' or 'B. Author, pers. comm.'). It is the authors' responsibility to obtain permission from colleagues to include their work as a personal communication. A letter of permission should accompany the manuscript.

The editor and publisher recommend that citation of online published papers and other material should be done via a DOI (digital object identifier), which all reputable online published material should have - see www.doi.org/ for more information. If an author cites anything which does not have a DOI they run the risk of the cited material not being traceable.

We recommend the use of a tool such as [EndNote](#) or [Reference Manager](#) for reference management and formatting. EndNote reference styles can be searched for here: www.endnote.com/support/enstyles.asp.

Reference Manager reference styles can be searched for here:www.refman.com/support/rmstyles.asp

5.4. Tables, Figures and Figure Legends

Figures and Tables: Always include a citation in the text for each figure and table. Artwork should be submitted online in electronic form. Detailed information on our digital illustration standards is available below. Any abbreviations used in figures and tables should be defined in a footnote.

Preparation of Electronic Figures for Publication: Print publication requires high quality images to prevent the final product being blurred or fuzzy. Submit EPS (line art) or TIFF (halftone/photographs) files only. MS PowerPoint and Word Graphics are unsuitable for printed pictures. Scans (TIFF only) should have a resolution of at least 300 dpi (halftone) or 600 to 1200 dpi (line drawings) in relation to the reproduction size (see below). Please submit the data for figures in black and white or submit a Colour Work Agreement Form (see Colour

Charges below). EPS files should be saved with fonts embedded (and with a TIFF preview if possible).

For scanned images, the scanning resolution (at final image size) should be as follows to ensure good reproduction: line art: >600 dpi; halftones (including gel photographs): >300 dpi; figures containing both halftone and line images: >600 dpi.

Further information can be obtained at Wiley-Blackwell's guidelines for figures: <http://authorservices.wiley.com/bauthor/illustration.asp>.

Check your electronic artwork before submitting
it: <http://authorservices.wiley.com/bauthor/eachecklist.asp>

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Section Two: Research Paper

**Exploring the Experiences of Being a Parent of a Child with Limb Difference who has
Been Provided with an Artificial limb**

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Doctorate in Clinical Psychology

Prepared in accordance with the journal guidelines for

Psychology and Health

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Abstract

Objectives: The study aimed to explore the experiences of parenting a child with limb difference who has been provided with an artificial limb. A unique aspect to this population is the negotiation of a prosthetic rehabilitation process but little research has explored this aspect of experience.

Design: Semi-structured interviews took place with seven parents. Interview data was recorded, transcribed and analysed using interpretative phenomenological analysis.

Results: The analysis identified four themes: (1) managing the initial emotional experience through the development of coping resources; (2) opportunities through prosthesis use and its relationship with 'normality'; (3) managing and making sense of social reactions toward their child; (4) the intrinsic role of support: developing a collective connection and enabling shared knowledge.

Conclusions: The study highlighted salient aspects to parents' experiences and sense-making that can inform clinical support. Emotional support, the management of social responses, and the holistic co-ordination of healthcare support with peer support networks are discussed. An interesting divergence around the experience and meaning related to their child's use of a prosthesis emerged concerning functionality. Healthcare professionals involved in the prosthetic rehabilitation process should look to explore these meanings to help support the management of the child's prosthesis use.

Key Words: Artificial Limb, Child, Limb Difference, Parenting, Prosthesis

Exploring the Experiences of Being a Parent of a Child with Limb Difference who has Been Provided with an Artificial limb

The term limb difference refers to an individual who has been born with a congenital limb deficiency or has acquired a limb loss during their life (Limbs4kids, 2015). Within the medical literature, limb differences are sometimes termed limb deficiencies and discussed in the context of a trauma, disease or a congenital condition (Dillingham, Pezzin, & Mackenzie, 2002; Makhoul, Goldstein, Smolkin, Avrahami, & Sujov, 2003). Limb difference in children is usually congenital in nature and occurs when part of, or the entire, limb does not form as expected during pregnancy (Vasluian, van Sluis et al., 2013). Complete epidemiological descriptions of limb difference in children are difficult to source and estimates can vary from country to country. Andrews, Williams, VandeCreek and Allen (2009) suggest prevalence rates for children born with a limb difference have tended to range between 3.5 to 7.1 in every 10,000 births. However, a recent 30-year population based study in northern Netherlands estimated congenital limb difference rates to be approximately 21.1 per 10,000 births (Vasluian, van Sluis et al., 2013); thus, suggesting inherent variability. Rates for acquired limb difference are equally hard to ascertain, and often occur through conditions such as cancer, severe health complications such as meningitis, or a physical trauma (Smith & Campbell, 2009). Children who have experienced a lower limb difference are almost always fitted with a prosthesis (artificial limb) to enhance their functional ability. Children with an upper limb difference will not necessarily always choose to wear a prosthesis as it will not always result in functional gain (Kuyper, Breedijk, Mulders, Post, & Prevo, 2001).

Psychosocial Impact on Children

Research exploring the psychosocial impact of limb difference on children and adolescents highlights variation within the research findings. For instance, Varni and Setoguchi (1992) reported an increased prevalence of emotional and behavioural issues and

lower social competence in children with acquired or congenital limb difference, compared to a normative community sample. In contrast, Hermansson, Eliasson and Engström (2005) found children with upper limb difference had similar levels of social competence and emotional and behavioural difficulties to a normative reference group. The authors did however find greater levels of withdrawn behaviour in children with limb difference.

In a review of the literature, Michielsen, Wijk and Ketelaar (2010) concluded the risk of psychosocial difficulties in children with limb difference to be comparable to children without a physical condition. The authors did comment on the methodological issues present in much of the research, which tends to employ cross-sectional designs. While cross-sectional studies can be useful to compare variables, they only provide a snapshot of a single moment in time; thereby, making it difficult to determine causation. Rumsey and Harcourt (2004) suggest the research literature exploring the impact of living with a congenital condition provides an inconsistent picture. They argue adjustment is likely to be a multi-faceted process involving individual, situational and societal factors, thus making generalisations difficult to capture.

Psychosocial Impact on Parents

There is a limited amount of research exploring the experiences of parents of children with limb difference. In a qualitative study looking at the response of parents following the birth of their child, Kerr and McIntosh (1998) documented parental feelings of shock, numbness and disbelief. In a subsequent study, Kerr and McIntosh (2000) found that many parents felt healthcare providers did not offer the necessary level of informational or emotional support. Parents reported a desire for information relating to both the aetiology and nature of their child's limb difference, and the likely development of their child. Parents also experienced feelings of isolation, worries related to social reactions, and concerns about the future. Building on this research, Andrews Williams, VandeCreek and Allen (2009) explored

parents' experiences of healthcare support and their perceived support needs during their child's first year of life. Using an open-ended questionnaire, the authors found parents consistently discussed their experiences in relation to three themes; attitudes, information and emotional support. This related to the attitudes healthcare providers possessed in relation to disability; the information and knowledge they have and share; and finally, the positive value they placed on the emotional support and compassion healthcare professionals provided. The authors suggest future research needs to continue to explore the impact of disability on parents and the potential mediating role of healthcare professionals.

The Role of Parental Adjustment

Research focusing on the role of parents and family functioning on a child's adjustment to a physical health condition is more substantial. Findings identify a reciprocal role between child adjustment and family functioning (Eiser, 1990), with chronic health conditions impacting, and impacted by, the functioning of families (Kazak, 1997; Wallander & Varni, 1998). Research has found parental adjustment and family support to be significant factors related to positive psychosocial adjustment in children with a chronic health or disfiguring condition (Bakker, Maertens, Van Son & Van Loey, 2013; Drotar, 1997; Perrin, Ayoub & Willett, 1993).

There have been a number of hypotheses in relation to the role of family functioning on child adjustment. For instance, parenting style has been shown to have a direct impact on child outcomes, with Morawska, Calam, and Fraser (2015) suggesting parents need to successfully integrate general parenting behaviour and specific health condition management. This can be difficult, particularly when a parent is unsure whether to make allowances for their child or enforce boundaries. Authoritative parenting practices, that is parenting with high levels of control and warmth, have been found to be associated with positive health related behaviours in children (Park & Walton-Moss, 2012). Parents' own stress levels have

also been hypothesised to influence their child's regulation of emotions (Klunnert, Kaugers, Strand & Silveira, 2008). This is important to consider as parenting a child with a disability has been found to result in increased parental stress (Baker-Ericzén, Brookman-Frazee, & Stahmner, 2005; Ylvén, Björck-Åkesson, & Granlund, 2006). Hall et al. (2012) suggest stress may occur through changes to the family system, with the effects potentially seen between parental relationships, the parent-child bond and relationships between siblings.

With reference to research exploring the role of parental adjustment in relation to children with limb difference, Varni and Setoguchi (1993) suggested paternal symptoms of anxiety and low mood, but not maternal, predicted child psychological adaptation. Family support was found to have a positive effect on child adaptation, as did social correlates of parent, classmate and friendship support. While the research focussed on individual parental differences, the findings highlight the positive role of the family on child adjustment.

Wallander and Varni (1992) developed a conceptual model of adjustment in children with chronic physical conditions that was informed by their research into paediatric limb loss and cancer. The model looks to conceptualize risk and resistance factors influencing ongoing child adjustment. The model refers to the role of the family environment and family member adjustment as a factor in promoting resilience, confirming the significant role of parents in paediatric health and psychosocial outcomes.

Prosthesis Use

The utilisation of a prosthesis by children has been researched in the context of functional outcomes, with some interesting differences between upper and lower limb prosthesis use. Ulger and Sener (2011) explored the use of a prosthesis amongst children with lower limb difference, both congenital and acquired. They found that after six months of prosthetic rehabilitation, the active use of a prosthesis could support functional gain.

Amongst children with upper limb difference, the use of a prosthesis is commonly rejected by

children due to lack of functional outcome. Parental disappointment in function, lack of involvement in treatment, and dissatisfaction regarding perceived support is associated with increased rejection rates (Postema, van der Donk, van Limbeek, Rijken, & Poelma, 1999).

Research exploring decisions to wear a prosthesis in children and adolescents can often be limited due to a reliance on fixed-response questionnaires. Recently however, Vasluian, de Jong, et al. (2013) used a qualitative framework approach to explore reasons for wearing or rejecting a prosthesis in young people with upper limb difference. Focus groups revealed that cosmesis was the prime factor in choosing to wear a prosthesis, with users feeling this enabled participation in social activities and led to less stares from others. Reasons for rejecting a prosthesis centred around weight and limited functionality. Interestingly, the authors suggested the participants who had come to accept their body image reported that their motivation for prosthesis use was not centred on the avoidance of social stares from others. This suggests interventions to enhance body image acceptance may interact with decisions to wear a prosthesis. Furthermore, parents' opinions regarding their child's prosthesis use were sought, with many motivated by a desire not to limit their child and give them the option of prosthesis use later in life. For parents of children who did not wear a prosthesis, the central issue was around functionality, and feeling that their child could function without one.

Interestingly, there appears to be a dearth of research exploring parents lived experiences of the prosthetic rehabilitation process. This comes somewhat as a surprise given the active involvement of the rehabilitation team in the management of prosthesis use (Coulter-O'Berry & Giavedoni, 2009). Of related interest, Smith and Campbell (2009) offered reflections based on their clinical experiences and discussed several key issues for parents regarding the utilisation of a prosthesis for their child. They discussed the importance of parenting style and discipline in negotiating issues around autonomy and independence

regarding their child's prosthesis use. This can be a difficult role for parents, with Smith and Campbell illuminating the emotional impact on parents. Additionally, research has also highlighted that the demands on parents extend to incorporate an economic burden due to frequent utilisation of healthcare appointments; parents often need to adjust their work schedules in response to their child's healthcare needs (Weir, Ephraim & Mackenzie, 2010).

The Current Study

In summary, parenting a child with limb difference has been associated with the experience of several strong emotions following the birth of their child. Research has also detailed dissatisfaction related to aspects of healthcare support, and worries related to perceived social reactions. The literature suggests parenting a child with a disability is a challenging role and can exacerbate parental stress. Given the significant role parents play in their child's adjustment, the present research is both timely and relevant.

A unique aspect to the experience of parenting a child with limb difference is the negotiation of the prosthetic rehabilitation process; however there appears to a void of research relating to this aspect of experience. Thus, the current study looks to address this gap in the literature using a phenomenological, qualitative approach which prioritises parents' lived experiences. The study intends to explore pertinent issues for parents, and in so doing make recommendations for effective support. This is harmonious with the assertion that child and family pathways of care should be built around the needs of those accessing them and to see "services through their eyes" (Department of Health, 2003, p.4). The primary research question was "What are the experiences of parents of a child with limb difference who have been provided with an artificial limb, and how do they make sense of these experiences?"

Method

Design

The study was concerned with prioritising parents' lived experiences and as such the utilisation of a qualitative research methodology was appropriate. A semi-structured interview was designed and utilised to elicit pertinent participant experiences, with the data analysed using interpretative phenomenological analysis (IPA). IPA is committed to exploring meaning and sense-making of a phenomenon amongst a well-defined sample (Smith & Osborn, 2008). It has its theoretical origins in phenomenology, idiography and hermeneutics: a focus on experience, the particular and interpretation (Smith, Flowers & Larkin, 2009). In practice the approach looks to explore how participants experience their world through the analysis of data from small, homogenous samples, acknowledging the active role of the researcher in the interpretation of these experiences. As Smith and Osborn (2008) report "the researcher is trying to make sense of the participants trying to make sense of their world" (p.53). The approach was chosen as it was congruent with the research question, which was looking to explore the experiences of parents of a child with limb difference and their ascribed meaning to these experiences.

Sampling and Participants

Consideration of the sample is an integral aspect to IPA studies. IPA takes an idiographic approach and so involves a detailed analysis moving from individual cases, to iteratively exploring convergences and divergences across participant accounts. Consequently, studies using IPA are typically conducted with small samples of participants that share or are bound by a particular experience. Smith and Osborn (2008) suggest sample size can be influenced by "the degree of commitment to the case study level of analysis, the richness of the individual cases, and the constraints one is operating under" (p. 56); typically published studies range from 4-10 participants. With these considerations in mind, a minimal sample of 4 and a maximum number of 12 was decided in advance. This was deemed both

satisfactory and pragmatic to explore the research question, while also respecting the detailed interpretative approach to analysis.

The criteria for inclusion in the study were parents of a child with a limb difference who had available to use a prosthesis for a minimum of six months before study participation. The child's age range for inclusion was between 5-16 years old. The rationale for this age range was to seek and capture parents' experiences relating to key cognitive, social and emotional developments for their child (e.g. having begun attending school, transition to high school, the developing identity), with the child's language development also a consideration. No restrictions were placed on the nature of the child's limb difference. Due to funding restrictions relating to the use of a translator, parents were required to converse in English. Parents of a child with a co-morbid health condition which may have limited or precluded prosthesis use were excluded, as were children with a co-morbid physical and/or intellectual disability. The rationale for both inclusion and exclusion criteria related to the preservation of homogeneity which is a fundamental aspect with IPA (Smith, et al., 2009).

Participants were recruited through the social media accounts of online charities supporting both children and adults with limb difference. Recruitment took place over a period of five months. A total of seven parents took part in the study, five parents resided in the United Kingdom, and two in the USA. Table 1 illustrates the key demographics of the included participants.

[INSERT TABLE 1]

Procedure

Participants who viewed the social media advertisement were invited to access an electronic version of the participant information sheet, and to contact the lead researcher if they were interested in taking part. Those participants who made contact were asked to fill an expression of interest form to ensure they met the inclusion criteria. Participants were invited

to express their preferences for the interview format, with a choice of Skype, telephone and face-to-face interviews offered. Due to the international nature of recruitment, the latter option was often not practical. Six parents chose to have their interview over the telephone, with one parent choosing face-to-face. At the end of the interviews participants were thanked for their time and provided with a debrief sheet containing information regarding supportive charity organisations.

Data Collection

A semi-structured interview schedule (see appendix A) was developed in collaboration with the research supervisor (who has published extensive research relating to amputation and prosthesis use), the field supervisor (a clinical psychologist with paediatric clinical experience) and liaison with a local limb difference charity. Questions looked to explore salient aspects of parents' experiences, including their perceived challenges related to the role and the experience of the prosthetic rehabilitation process. The aim was to produce a schedule that would suitably explore the research question, but be flexible enough to enable participants to direct and disclose aspects of their experience relevant to the aims of the research. Interview length ranged from 47 minutes to 66 minutes, with an average length of 57 minutes. All interviews were then transcribed verbatim by the lead researcher, with pseudonyms generated to maintain confidentiality.

Data Analysis

Smith and Osborn (2008) suggest IPA is both an epistemological and methodological approach that does not have a prescriptive process to data analysis. The primary aim of the analysis can be characterised by moving from the particular to the shared, from the descriptive to the interpretative (Smith et al., 2009). Participant transcripts were analysed one at a time to retain IPA's idiographic focus (Snelgrove, 2014). Firstly, transcripts were read several times, with the audio recording also played to aid familiarisation. Initial codes were

then generated for each transcript (appendix B). These codes captured the experiences and sense making aspects of participants' responses that related to the research question. These codes were then iteratively grouped together into discrete groups that shared aspects of experience. In accordance with the guidelines of Murray and Wilde (in press), an interpretative narrative summary was then written for each group of codes that captured the salient experiences and meanings of the participant's account. A title was then given that looked to encapsulate the interpretative narrative; thereby naming the idiographic themes for each participant (appendix C). Once this process was completed for each participant, themes were integrated across participants, noting convergences and divergences, with a synthesised narrative summary produced for each cluster of themes. This led to the production of final themes capturing a more generalised understanding of parental experiences and sense making.

Credibility of Analysis

Yardley (2000) discusses some of the issues and dilemmas regarding the assessment of quality in qualitative research, which is not always easy to define due to the inherent complexity within the paradigm. The article however does predicate the importance of thoroughness in data collection, analysis and rigor. In accordance with these recommendations, a thorough audit trail relating to the analysis of each participant was completed (see example appendix D). In addition, collaboration with the research supervisor was a central aspect relating to the rigor of the analysis. Initial coding excerpts from anonymised transcripts and the audit trail illustrating the generation of idiographic themes, were sent to the research supervisor after the analysis of the first participant. Feedback was provided concerning the level of interpretation in the analysis. For example, the narrative summary in Lisa's account relating to the theme 'Making Sense of the Initial Shock: Adjusting and Adapting to a New Idea of Parenthood' initially lacked interpretation relating

to how Lisa's experiences impacted on her identity as a parent. Furthermore, each theme is also evidenced by extracts from between five and seven participants which exceeds Smith's (2011) 'acceptable' criteria when assessing the quality in IPA studies.

Reflexivity

IPA acknowledges the role of the researcher in the research process through a process of *double hermeneutics*; this is the acknowledgement that participants are trying to make sense of their own experiences, which in turn are then interpreted by the researcher. Thus, it is important for the researcher to adopt a position of reflexivity and attempt to bracket personal beliefs and assumptions (Dahlberg, 2006; Starks & Trinidad, 2007). During the data collection period, the lead researcher was completing a Doctorate in Clinical Psychology. To help develop a sense of self-awareness in the research process, the lead researcher made a brief note before and after interviews to acknowledge any thoughts, feelings or actions that may have influenced the data generated. For example, after the first telephone interview, the lead researcher was left to reflect on the importance of active listening skills, particularly the use of reflecting back participant responses to increase engagement in the absence of any non-verbal cues.

Ethical Review

The research was given ethical approval by Lancaster University's ethics committee following the development of a research protocol. Professional guidelines from the British Psychological Society (2014) and the Health and Care Professionals Council (2015) were considered throughout. Central to this was a consideration of managing ethical issues related to the use of mediated communication methods. Informed consent was ensured through access to an electronic participant information sheet, and an electronic consent form shared at least 24 hours before any interview took place; verbal consent was recorded (see ethics section p.30-32). Finally, due to the nature of telephone interviews, due consideration of

safeguarding and risk issues was essential. A safety plan was developed and agreed with both the research and field supervisor. A debrief sheet (ethics section p.37) was also developed and supplied details of a worldwide organisation providing emotional support. All participants were offered a full copy and/or summary of the research paper on its conclusion.

Results

The analysis led to the generation of four main themes: (1) managing the initial emotional experience through the development of coping resources; (2) opportunities through prosthesis use and its relationship with 'normality'; (3) managing and making sense of social reactions toward their child; and (4) the intrinsic role of support: developing a collective connection and enabling shared knowledge. Table 2 illustrates the development of the final themes from individual participant themes.

[INSERT TABLE 2]

Theme 1. Managing the Initial Emotional Experience Through the Development of Coping Resources

All parents in the study recalled strong feelings such as shock when they found out about their child's limb difference. There was a sense that this challenged their pre-existing vision of being a parent: "When you are pregnant you think about your child and imagine them doing things like going to brownies or riding a bike or going to school and then obviously, you suddenly think how is she going to do that" (Lisa).

There was a feeling of uncertainty about the challenges that lay ahead, with most parents having little knowledge of limb difference before becoming a parent:

I suppose it was a challenge to kind of get our heads around his disability when we first found out, and to learn about what it would mean and the impact it would have on his life, and our lives. (Francis)

Many parents discussed a perceived need to protect their child and to provide the necessary conditions to ensure the best outcome. For some, this necessitated a desire to put their child's needs above their own. Patricia explained: "I think I have always done mind over matter and tried to get on with it"; here the metaphor was used to illustrate how she puts her own needs to one side to prioritise her child's.

In response to the initial emotional experience, parents seemed to develop their own coping resources. These coping resources seemed to help parents to both manage and make sense of the initial emotional experience and the perceived challenges that lay ahead. For instance, Elizabeth and her husband decided not to let limb difference limit their child, and discussed coping through the development of a positive attitude and a proactive approach to overcoming barriers: "We both, my husband and I, we don't really let it be an excuse in his life". For some parents, there was a sense of advocacy in the way they coped, which seemed to be in response to a perceived duty to protect the needs of their child. Jill reflected on her approach to overcoming barriers related to prosthesis provision: "so it is just really just advocating". For Lisa, her sense of advocacy was translated into a perceived need to anticipate future challenges: "I've always tried to be one step ahead". There was a sense of an organic nature to the coping resources developed, with their ways of coping evolving as their child gets older and in response to ongoing challenges.

Finally, within some accounts there was a sense of reflection regarding their initial concerns and fears not being realised, and descriptions related to the positive experiences of parenting a child with limb difference: "It is nowhere near as bad as you paint it to be in your nightmares. And it could actually turn out to be in some weird way to be a blessing or positive experience for all of you" (Clare).

In summary, the theme encapsulates the initial emotional experience for parents, which was a difficult period as they tried to adapt to the perceived challenges ahead. In

response to these initial feelings parents seemed to develop coping resources to ensure they could fulfil their perceived parental responsibilities, with these coping resources evolving to meet ongoing demands.

Theme 2. Opportunities Through Prosthesis Use and its Relationship with ‘Normality’

Their child’s use of a prosthesis was a central part of parents’ experiences. For most it appeared to enable opportunities for their child that they did not envisage when they discovered their child’s limb difference. It allowed them to experience activities alongside their peers: “So if he didn’t have the leg he has, he wouldn’t be able to ride a bike, he plays football, he does skiing, any of that, he wouldn’t be able to do” (Patricia). Jill, whose child plays competitive sport, reflected how the use of a prosthesis vindicated the decision to amputate: “I knew that amputation was the best for her because of prosthetics. She would never have done what she has done if we did not choose to amputate, she would be disabled”.

Through parents’ accounts, prosthesis use seemed to relate to a sense of normality, where their child could engage in activities like their peers and be treated like their peers. Clare discussed how her child’s prosthesis use has enabled a “normal” family life:

It’s been a normal life, like I worry so much more about like him getting in trouble at school, you know underachieving, him cracking his head open, him falling out of a tree, not being kind to his sister. All these things are much more and part of my mind on a daily basis than the fact he has a prosthetic leg, it doesn’t even cross my mind, because it has zero impact on the quality of his life. (Clare)

Parents discussed the use of a prosthesis as contributing to a sense of confidence and enhanced self-esteem for their child. Patricia discussed a sense of pride in how her child had adapted to their prosthesis and acknowledged this had made things easier for her: “So the fact that he has just got on with it and doesn’t see himself as any different from anyone else, makes it easier for me”.

Conversely, Lisa and Francis, whose children have upper limb difference, were more ambivalent about prosthesis use. Francis critically considered the function of a prosthesis. She felt its use was mainly for cosmetic reasons and made her child feel more different to their peers because of its limited functionality:

It became very apparent that it was completely unnecessary, [child] didn't particularly take to it, he didn't like it, he just used to pull it off. We were not that enthusiastic either because it just seemed so rudimentary and not really that helpful. (Francis)

Similarly, Lisa commented: "She can do more without it" but commented her child will wear it to avoid social stares. The lack of function seemed to lead Francis to reflect on societal attitudes toward 'normality':

I think more and more, kind of attitudes to disabled people are changing and it is not a case of sticking a prosthetic on and making people look the same and being 'normal' and two handed, people are more accepting of people for who they are so I think they [prostheses] seem to be less popular nowadays. (Francis)

In summary, parents conveyed how the utilisation of a prosthesis enabled opportunities for their children and participation in activities alongside their peers. For some parents, there was a sense that using a prosthesis allowed their child to be treated the same as their peers and enabled a sense of normality. However, there appeared to be a divergence within the account, with this aspect of experience seemingly not central to parents of children with upper limb loss, where there appeared a critical appraisal regarding its functionality.

Theme 3. Managing and Making Sense of Social Reactions Toward Their Child

All parents discussed the challenge of social reactions toward their child and how they managed and made sense of them. For some, this challenge provided a direct conflict and tension to a parental instinct to protect their child. Francis recalled an incident at her child's school where they were not included because of their limb difference: "It is only isolated

incidents but that upsets him and you obviously have then got to be strong and supportive to him but obviously, your heart is breaking inside as well”.

There was a sense of disenchantment regarding members of the public staring: “I think it can be disappointing. I think you are just disappointed in humanity sometimes, it’s frustrating” (Clare). The impact of social reactions seemed to agitate parents’ own beliefs that their child should not be treated differently from others: “Don’t stare at them or make them feel under pressure, or make them feel as if everybody is looking at them thinking that they are different” (Patricia).

Parents discussed managing the impact of social stares by attributing them to a natural curiosity to difference, Ruth explained: “You get a lot of kids staring and looking just out of curiosity, I think mostly that is what it is, it is not nastiness, its human nature to look at something that is a bit different”. Similarly, Clare commented: “Often people aren’t staring in a negative way, they are staring in kind of admiration, so we just try to talk to him a lot about it”. Jill discussed modelling to her child that others will stare and want to ask questions about their leg. She felt it was important for her child “not to get frustrated and not to think they [others] are mean”. She felt parents’ responses to social reactions were important to consider and would “dictate how your child feels later in life”.

The experience of social responses left some parents to reflect on societal attitudes toward disability. Some parents took comfort in a greater media profile of disability, with the Paralympics cited as a source of positive visibility. For some, this visibility translated into more inclusive societal attitudes: “I think there has been a huge rise in the acceptance and the awareness of disabilities” (Ruth). Francis added: “Just having more people visible in the media and on the television and stuff is really a source of support for the parents and for our children”. Additionally, Francis also felt her child’s classmates would develop a greater

appreciation of issues related to difference: “I think it is positive to other children as well because it sends a message to them that people are different”.

In summary, social responses created a considerable challenge to parents. The experience of negative social responses was difficult for parents and seemed to relate to a desire to protect their child and a belief that they should not be treated differently. There was an aspect of experiencing social reactions that seemed to lead to wider reflections around societal attitudes toward difference, with some parents taking solace from increased media visibility.

Theme 4. The Intrinsic Role of Support: Developing a Collective Connection and Enabling Shared Knowledge

The importance of feeling supported was emphasised in all parent accounts. The support provided parents with a shared connection and a shared understanding of their parenting role. Ruth encapsulated the value of parent-to-parent support, which related to a sense of emotional containment and validation:

I think it is just a comfort blanket, knowing that if you ever have a real fear or a real feeling of anxiety or whatever it happens to be, there is somebody that you can [talk to], you don't have to keep it in anymore you can let it out. (Ruth)

There was also a benefit of developing a support network that related to a pragmatic benefit of staying connected and knowledgeable in relation to prosthesis provision. This seemed to relate to parents' initial feelings of uncertainty and the unknowing aspect to becoming a parent of a child with limb difference. Jill referred to her “huge support network” and felt an important aspect of being a parent of a child with limb difference was to stay connected. Lisa discussed how a charity social media account helped her to become aware of what was available through the healthcare service in terms of prosthesis provision: “When she started going down the myo-electric journey, the initial ones she had were not brilliant

and through a lad I know through [charity], I knew he got one of the latest model ones from a different limb centre”.

Some parents discussed the benefit of peer support in the form of family events or camps for children with limb difference. These events seemed to offer a sense of normalisation of their experiences and a feeling of acceptance:

There are just loads of kids like [child] and parents like us, you know, sometimes you don't even talk about limb differences, you can just sort of sit and know that your child isn't going to be stared at, they are going to be accepted and that everyone is really positive. (Francis)

The importance of feeling supported extended to healthcare providers. Parents valued the relationship with their prosthetist, which related to a sense of trust, and the prosthetist wanting the best outcome for their child: “He was clearly wanting the best for [child]. It isn't just a job to him” (Clare). However, healthcare support was not always appraised as positive. Parents could experience a lack of emotional support in the immediate aftermath of finding out about their child's limb difference. There was a sense this was a disappointing aspect of care given the important healthcare decisions parents often had to make around this time. Ruth discussed her experience of support when deciding whether to have her child's leg amputated: “I think there should have been something, especially when you are getting told the darkest news that her leg is not going to grow and there is basically nothing we can do about it”.

In summary, developing support networks was highly valued and offered both emotional and practical support. The support offered by healthcare services and professionals was also a prominent narrative in parent accounts. Parents offered a collective regard to the development of a trusting relationship with their prosthetist. Many parents discussed a lack of emotional support around the time of their child's diagnosis, with a sense this was a

disappointing aspect of care received given the emotional challenges they faced during this period.

Discussion

The aim of this study was to explore and make sense of the experiences of parenting a child with limb difference who has been provided with an artificial limb. Using a semi-structured interview with seven participants the results produced four main themes. The themes will be discussed in relation to existing research, with discussion around the significant issues and clinical implications offered.

The results suggest that upon finding out about their child's limb difference, whether during pregnancy or birth, parents experience a range of powerful emotions, supporting previous research (Kerr & McIntosh, 1998). Within parent accounts there was a sense of uncertainty about the challenges that lay ahead and a move away from their pre-existing ideas of being a parent. In response to their initial emotional experience, parents developed a variety of coping resources to help them make sense of and manage the perceived challenges ahead. This is in keeping with the resiliency model of family stress, adjustment and adaptation (McCubbin & McCubbin, 1993), which conceptualises the process of adaptation and resiliency in families as an ongoing, evolving process. The model looks at the interaction of family responses to a stressor (child's limb difference), paying attention to their vulnerability at the time of the stressor, their functioning patterns and resistance resources, and their appraisal of the stressor. In response to these interactions, the family use problem solving or coping strategies to look to manage the demands. The model is essentially looking to build on existing resources within the family and use them to inform effective coping strategies to maintain emotional stability (Frain et al., 2007).

The role of social support, particularly parent-to-parent support was valued by parents in the present study. The positive influence of social support is a consistent presence within

paediatric models of adjustment (e.g. Thompson, Gil, Burbach, Keith, & Kinney; Wallander & Varni, 1992) and falls within the resistance resources in the resiliency model of family stress, adjustment and adaptation (McCubbin & McCubbin, 1993). All interviewed parents commented on the value of support in the form of other parents of children with limb difference. The value of parent-to-parent support was also highlighted in Vasluian, de Jong et al.'s (2013) study. Interestingly, the results of the present study extended parents' perceived value of support to include a more pragmatic aspect of information sharing. Support networks enabled parents to educate themselves around important issues related to their child's limb difference such as prosthesis provision.

The results also highlighted the experience of social reactions for parents. For some parents, there was a sense of disillusionment regarding members of the public staring at their child, with others choosing to take meaning from this experience in relation to a normal human propensity to look at something that is different. Parents worried about the impact of social stares on their child's self-esteem and confidence: research suggests this is a valid concern (Rumsey & Harcourt, 2007). The experience of social responses seemed to lead some parents to reflect more generally about societal attitudes toward disability, with many feeling society is becoming more tolerant and mindful of issues related to inclusivity. Increased media coverage of disability was a source of support for many parents, with some citing the Paralympic Games as a positive example. This seemed to give individuals with a disability a platform to showcase their abilities and normalise the use of a prosthesis. This finding supports Gunter's (2012) assertion regarding the educative power of the media, and the widely held belief it can help promote diversity and challenge appearance stereotypes.

A central feature of the experience of parents concerned the meanings attached to prosthesis use for their child; with this aspect of experience novel to the research literature. The results suggested that for most parents, their child's use of a prosthesis enabled

opportunities, social integration and for them to be treated the same as their peers. This finding echoes the work of Murray (2010) who focussed on adult users of a prosthesis and the personal and social meanings attached to its use. Murray suggested the personal meaning can go beyond the functional capabilities of the device, and be entangled with more significant meanings, such as the prosthesis enabling the user to feel like they are living a 'normal' life, being treated the same as non-users, and enabling independence and social participation.

An interesting divergence was found between upper limb prosthesis use and lower limb prosthesis use, with the former appearing to take a more overt critical stance regarding its functionality. The issue of functionality has received attention before, with Postema et al. (1999) suggesting lack of function is often a pivotal reason for the rejection of upper limb prosthetics by both children and their parents. Lower limb prosthesis use seemed to enable function comparable to same aged peers, and for parents this allowed their child to be treated the same. This was less transparent amongst parent accounts of their child's upper limb prosthesis use, who were ambivalent regarding its function and use more generally. Interestingly, Vasluian, de Jong, et al. (2013) found that some older adolescents suggested they decided not to use an upper limb prosthesis as it made them feel more disabled; this is in line with Francis' report of her own child's beliefs regarding prosthesis use.

Thinking more broadly about the results of the study, it is of note that many of the themes did not directly relate to prosthesis use, and may best be understood as indirectly related to prosthesis use (for instance the experience of social stares). The experiences and meanings parents described may resonate with parents of children with other health conditions, particularly children with a visible difference. A central tenet of IPA is to privilege the meanings and understandings of participants. Therefore, it is essential to stay close to the participants own words during the interview and not allow the researcher's ideas and understandings to dictate it (Murray & Wilde, in press). The results of the present study

therefore reflect the experiences and meanings that were most prominent within parents' accounts.

Clinical Implications

The results have a number of clinical implications that can help inform effective care for both children and parents of children with limb difference. Firstly, there is a need for service provision to show a greater consideration of parental emotional support. In the UK, hospital services adopt a child-centred approach to care that should consider the support needs of parents to help them understand and cope with their child's health condition (Department of Health, 2003). However, the results of the present study suggest a greater focus on the emotional needs of parents should be emphasised.

Supporting parents seems to make intuitive sense with research consistently illustrating a reciprocal role between child and parent adjustment in children with health conditions (e.g. Thompson et al. 1993). Mercer et al. (2015) recently discussed the delivery of psychological support for children and their families within paediatric settings. They advocate for the need to identify and support parents who may be finding it difficult to cope, as this will ultimately impact their own child's ability to cope. Furthermore, they suggest a role for clinical psychologists to be embedded within multi-disciplinary teams (MDT) and use their skills in consultation, joint-working, training and supervision. This can help to promote issues related to psychological well-being as being everyone's responsibility within an MDT.

Healthcare services are now considered to adopt a model of patient-centred care which includes shared decision making between provider and patient (Dwamena et al., 2012). However shared decision making is not always easy to navigate. Research suggests decision making around treatment options by parents in healthcare settings can be influenced by a moral desire to 'do the right thing' and enhance social inclusion for their child (Nelson,

Caress, Glenny & Kirk, 2012). Supporting parents to consider their feelings and personal meanings attached to prosthesis use for their child, and issues related to cosmesis and functionality, may help parents to feel more confident in managing their child's use of a prosthesis. This may lend itself well to the use of formulation by a clinical psychologist (Division of Clinical Psychology, 2011). Given the central role parents play in the decisions and management of their child's prosthesis use, the results advocate for parents' meanings of prosthesis use to be explored when engaging within the prosthetic rehabilitation process.

Parents' experiences of social reactions toward their child, and the ongoing challenge this presents should also be carefully considered. Social learning theory (Bandura, 1977) can be a helpful model to understand how behaviour can be modelled; specifically, to consider how parents' reaction to social stares may be modelled to their child. Parents may benefit from exploring their own opinions and views around issues related to visible difference, with some parents reflecting on a change in their own attitudes toward difference. Research suggests a parent's feelings about their child's appearance can be assimilated and internalised by the child, which in turn can influence their own conceptions of body image and feelings of self-worth (Kearney-Cooke, 2002). Therefore, exploring this with parents in a non-judgemental and non-stigmatizing way may be of value.

A key clinical implication concerns the co-ordination of healthcare services with relevant third sector voluntary agencies to ensure parent-to-parent support networks can be developed. In addition to the emotional support parents gained from a network of peer support, there was also a practical or pragmatic aspect that related to information sharing and prosthesis provision. Parents perceived lack of information fits with prior research findings (Kerr & McIntosh, 2000), and suggests further work need to be done regarding information sharing by healthcare professionals around the time of diagnosis and the birth of their child.

Finally, it seems important to note that many of the experiences parents discussed were not specifically related to prosthesis use. Parent's initial emotional experience, the experience of social responses toward their child, and the importance of both peer and professional support were all prominent aspects of parent experience. Healthcare professionals and prosthesis services supporting parents and their child with limb difference may benefit from reflecting on some of these wider issues when thinking about effective support.

Limitations

In consideration of the sample, the absence of any fathers in the present research is a limitation. It is conceivable that the results do not portray salient aspects of experience that are relevant to the experiences of fathers. For instance, Jill indicated her husband responds differently to social stares toward their child, which may have added to the dimensions relating to how parents manage and make sense of social responses. The absence of fathers within parenting research remains an ongoing issue (Phares, Lopez, Fields, Kamboukos, & Duhig, 2005). In addition, only one parent in the study had a child with an acquired limb difference, therefore the results may not represent the salient experiences and sense-making relevant to this group of parents.

The recruitment method of using the social media accounts of charities could also present a problem in that it may fail to represent parents who have not drawn on the support of others, or even purposefully avoided peer support. However, one benefit of the recruitment method is that it obtained an international sample, thereby obtaining a diverse set of experiences while retaining homogeneity.

Finally, this research was exploratory in nature, using a small but detailed account of parental experiences. It is therefore important to acknowledge that the results are not attempting to generalise to the experiences of all parents of children with limb difference.

Smith et al. (2009) suggest IPA studies can offer theoretical generalisability, which gives the reader the opportunity to assess the results in the context of their professional and experiential knowledge.

Further Research

The research identified an interesting divergence in parental accounts regarding the functionality of upper and lower limb prosthesis use. Future research could explore this further and prioritise both parent and their child's personal and social meanings attached to prosthesis use, in a similar way to the work of Murray (2010) with adult prosthesis users. Exploring meanings attached to prosthesis use across a variety of ages would be beneficial, for instance in adolescence, with Erikson's (1968) psychosocial model of development suggesting this period to be associated with identity formation. Exploring this in relation to prosthesis use could help support rehabilitation teams to provide effective, service-user informed support.

The present study did not differentiate between acquired and congenital limb difference. While this did not impact the results of the present study, with one parent having a child who acquired their limb difference due to a surgical complication at a young age, future research could explore this further and detail any aspects of experiences that may differ. Once more this could have implications in providing effective support for parents.

Conclusion

The findings suggest that parents experience a range of strong emotions as they adjust to their child's diagnosis of limb difference, with coping resources developed in response to the perceived challenges ahead. For most parents their child's use of a prosthesis enabled participation opportunities and facilitated their child being treated like their peers. However, for parents of children using an upper limb prosthesis there was a more critical appraisal regarding functionality. The ongoing challenge presented by social responses was

highlighted, with parents looking to protect their child from any negative impact. Parents drew support from media visibility and a perceived attitudinal shift toward inclusivity. The value of both peer and healthcare support cannot be understated, with services seemingly requiring a greater focus on providing emotional support to parents, as well as a more coordinated relationship to peer support networks.

References

- Andrews, E. E., Williams, J. L., Vandecreek, L., & Allen, J. B. (2009). Experiences of parents of children with congenital limb differences with health care providers: a qualitative study. *Rehabilitation Psychology, 54*(2), 217-221. doi:10.1037/a0015651
- Baker-Ericzén, M. J., Brookman-Fraee, L., & Stahmner, A. (2005). Stress levels and adaptability in parents of toddlers with and without autism spectrum disorders. *Research & Practice for Persons with Severe Disabilities, 30*(4), 194–204. doi:10.2511/rpsd.30.4.194
- Bakker, A., Maertens, K. P., Van Son, M. M., & Van Loey, N. E. (2013). Psychological consequences of pediatric burns from a child and family perspective: a review of the empirical literature. *Clinical Psychology Review, 33*(3), 361-371. doi:10.1016/j.cpr.2012.12.006
- Bandura, A. (1977). *Social learning theory*. Englewood Cliffs, NJ: Prentice Hall.
- British Psychological Society. (2014). *Code of human research ethics*. Retrieved from http://www.bps.org.uk/system/files/Public%20files/code_of_human_research_ethics_dec_2014_inf180_web.pdf
- Coulter-O'Berry, C., & Giavedoni, B. (2009). Rehabilitation of the child with lower limb loss. *Journal Of Pediatric Rehabilitation Medicine, 2*(3), 159-172. doi:10.3233/PRM-2009-0088
- Dahlberg, K. (2006). The essence of essences – the search for meaning structures in phenomenological analysis of lifeworld phenomena. *International Journal Of Qualitative Studies On Health & Well-Being, 1*(1), 11-19. doi:10.1080/17482620500478405
- Department of Health. (2003). *Getting the right start: National service framework for children standard for hospital service*. London: The Stationary Office.

- Dillingham, T. R., Pezzin, L. E., & Mackenzie, E. J. (2002). Limb amputation and limb deficiency: Epidemiology and recent trends in the United States. *Southern Medical Journal*, 95, 875–883. doi: 10.1097/00007611-200208000-00018
- Division of Clinical Psychology. (2011). *Good practice guidelines on the use of psychological formulation*. Leicester: British Psychological Society.
- Drotar, D. (1997). Relating parent & family functioning to the psychological adjustment of children with chronic health conditions: What have we learned? What do we need to know? *Journal Of Pediatric Psychology*, 22(2), 149-165. doi:10.1093/jpepsy/22.2.149
- Dwamena, F., Holmes-Rovner, M., Gaulden, C. M., Jorgenson, S., Sadigh, G., Sikorskii, A., & ... Olomu, A. (2012). Interventions for providers to promote a patient-centred approach in clinical consultations. *The Cochrane Database Of Systematic Reviews*, 12CD003267. doi:10.1002/14651858.CD003267.pub2
- Eiser, C. (1990). Psychological effects of chronic disease. *Journal of Child Psychology and Psychiatry, and Allied Disciplines*, 31(1), 85-98. doi:10.1111/j.1469-7610.1990.tb02274.x
- Erikson, E. (1968). *Identity, youth and crisis*. New York: Norton.
- Frain, M. P., Berven, N. L., Tschopp, M. K., Lee, G. K., Tansey, T., & Chronister, J. (2007). Use of the resiliency model of family Stress, adjustment and adaptation by rehabilitation counselors. *Journal Of Rehabilitation*, 73(3), 18-25. Retrieved from <https://www.ebscohost.com/academic/amed-the-allied-and-complementary-medicine-database>
- Gunter, B. (2012). The role of the media. In N. Rumsey & D. Harcourt (Eds.), *The oxford handbook of the psychology of appearance* (pp 455-467). Oxford: Oxford University Press.

Hall, H. R., Neely-Barnes, S. L., Graff, J. C., Krcek, T. E., Roberts, R. J., & Hankins, J. S.

(2012). Parental stress in families of children with a genetic disorder/disability and the resiliency model of family stress, adjustment, and adaptation. *Issues in*

Comprehensive Pediatric Nursing, 35(1), 24-44. doi:10.3109/01460862.2012.646479

Health and Care Professionals Council. (2015). *Standards of conduct, performance and ethics*. Retrieved from

http://www.hpcuk.org/assets/documents/10002963sop_practitioner_psychologists.pdf

Hermansson, L., Eliasson, A., & Engström, I. (2005). Psychosocial adjustment in Swedish

children with upper-limb reduction deficiency and a myoelectric prosthetic hand. *Acta*

Paediatrica, 94(4), 479-488. doi:10.1080/0803525041002251

Kazak, A. E. (1997). A contextual family/systems approach to pediatric psychology:

Introduction to the special issue. *Journal of Pediatric Psychology*, 22(2), 141-148.

doi:10.1093/jpepsy/22.2.141

Kearney-Cooke, A. (2002). Familial influences on body image development. In T. F. Cash &

T. Pruzinsky (Eds.), *Body image: A handbook of theory, and clinical practice* (pp. 99–107). New York: Guilford.

Klennert, M. D., Kaugars, A. S., Strand, M., & Silveira, L. (2008). Family psychological

factors in relation to children's asthma status and behavioral adjustment at age 4.

Family Process, 47(1), 41-61. doi:10.1111/j.1545-5300.2008.00238

Kerr, S. M., & McIntosh, J. B. (1998). Disclosure of disability: exploring the perspective of

parents. *Midwifery*, 14(4), 225-232. doi:10.1016/S0266-6138(98)90094-8

Kerr, S. M., & McIntosh, J. B. (2000). Coping when a child has a disability: Exploring the

impact of parent-to-parent support. *Child: Care, Health and Development*, 26, 309–

322. doi:10.1046/j.1365-2214.2000.00149.x

- Kuyper, M., Breedijk, M., Mulders, A., Post, M., & Prevo, A. (2001). Prosthetic management of children in the Netherlands with upper limb deficiencies. *Prosthetics and Orthotics International*, 25(3), 228. doi:10.1080/03093640108726606
- Limbs4kids. (2015). *Fact Sheet 1: About limb difference and limb difference professionals*. Retrieved from: <http://limbs4kids.org.au/wp-content/uploads/2015/05/Fact-Sheet-1-About-LimbDifference.pdf>
- Makhoul, I. R., Goldstein, I., Smolkin, T., Avrahami, R., & Sujov, P. (2003). Congenital limb deficiencies in newborn infants: Prevalence, characteristics and prenatal diagnosis. *Prenatal Diagnosis*, 23, 198–200. doi: 10.1002/pd.550
- McCubbin, M. A., & McCubbin, H. I. (1993). Families coping with illness: The resiliency model of family stress, adjustment, and adaptation. In C. B. Danielson, B. Hamel-Bissell, & P. Winstead-Fry (Eds.), *Families, health, and illness: Perspectives on coping and intervention* (pp. 21–63). St. Louis, MO: C.V. Mosby.
- Mercer, A., O'Curry, S., Donnan, J., Stedmon, J., Reed, J., & Griggs, H. (2015). Delivering psychological services for children and young people with physical health needs and their families. *Child & Family Clinical Psychology Review*, 3, 71-81.
- Michielsen, A., van Wijk, I., & Ketelaar, M. (2010). Participation and quality of life in children and adolescents with congenital limb deficiencies: A narrative review. *Prosthetics and Orthotics International*, 34(4), 351-361. doi:10.3109/03093646.2010.495371
- Morawska, A., Calam, R., & Fraser, J. (2015). Parenting interventions for childhood chronic illness: A review and recommendations for intervention design and delivery. *Journal of Child Health Care*, 19(1), 5-17. doi:10.1177/1367493513496664
- Murray, C. (2010). Understanding adjustment and coping to limb loss and absence through phenomenologies of prosthesis use. In C. Murray (Eds.), *Amputation, prosthesis use,*

and phantom limb pain. An interdisciplinary perspective (pp. 81-100). London:

Springer

Murray, C., & Wilde, D. J. (in press). Phenomenology and interpretative phenomenological analysis. In S. Cartwright, C. Walshe, & S. Brearley (Eds.), *Handbook of theory and methods in applied health research*. Cheltenham: Edward Elgar.

Nelson, P. A., Caress, A., Glenney, A., & Kirk, S. A. (2012). 'Doing the "right" thing': how parents experience and manage decision-making for children's 'normalising' surgeries. *Social Science & Medicine*, 74(5), 796-804. doi:10.1016/j.socscimed.2011.11.024

Park, H., & Walton-Moss, B. (2012). Parenting style, parenting stress, and children's health-related behaviors. *Journal of Developmental And Behavioral Pediatrics* 33(6), 495-503. doi:10.1097/DBP.0b013e318258bdb8

Perrin, E. C., Ayoub, C. C., & Willett, J. B. (1993). In the eyes of the beholder: Family and maternal influences on perceptions of adjustment of children with a chronic illness. *Journal Of Developmental And Behavioral Pediatrics*, 14(2), 94-105. doi:10.1097/00004703-199304000-00004

Phares, V., Lopez, E., Fields, S., Kamboukos, D., & Duhig, A. M. (2005). Are fathers involved in pediatric psychology research and treatment? *Journal of Pediatric Psychology*, 30(8), 631-643. doi:10.1093/jpepsy/jsi050

Postema, K., van der Donk, V., van Limbeek, J., Rijken, R., & Poelma, M. (1999). Prosthesis rejection in children with a unilateral congenital arm defect. *Clinical Rehabilitation*, 13(3), 243-249. doi:10.1177/026921559901300308

Rumsey, N., & Harcourt, D. (2004). Body image and disfigurement: issues and interventions. *Body Image*, 1(1), 83-97. doi:10.1016/S1740-1445(03)00005-6

Rumsey, N. & Harcourt, D. (2007). Visible difference amongst children and adolescents:

Issues and interventions. *Developmental Neurorehabilitation*, 10, 113-123.

doi:10.1080/13638490701217396

Smith, D. G. & Campbell, K. M. (2009). Prosthesis for children and teens with limb

difference: Issues and expectations. *InMotion*, 19(2) 12-18. Retrieved from

http://www.amputecoalition.org/inmotion/mar_apr_09/prostheses_for_children.html

Smith, J. A. (2011). Evaluating the contribution of interpretative phenomenological analysis.

Health Psychology Review, 5(1), 9-27. doi:10.1080/17437199.2010.510659

Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative phenomenological analysis:*

Theory, method and research. London: Sage

Smith, J. A., & Osborn, M. (2008). Interpretative phenomenological analysis. In S. A. Smith

(Eds.) *Qualitative psychology: a practical guide to methods* (pp. 53-80). London:

Sage.

Snelgrove, S. R. (2014). Conducting qualitative longitudinal research using interpretative

phenomenological analysis. *Nurse Researcher*, 22(1), 20-25.

doi:10.7748/nr.22.1.20.e1277

Starks, H., & Trinidad, S. B. (2007). Choose your method: A comparison of phenomenology,

discourse analysis, and grounded theory. *Qualitative Health Research*, 17(10), 1372-

1380. doi:10.1177/1049732307307031

Thompson, R. J., Gil, K. M., Burbach, D. J., Keith, B. R., & Kinney, T. R. (1993). Role of

child and maternal processes in the psychological adjustment of children with sickle

cell disease. *Journal of Consulting and Clinical Psychology*, 61,

468 – 474. doi:10.1037/0022-006X.61.3.468

- Ulger, O., & Sener, G. (2011). Functional outcome after prosthetic rehabilitation of children with acquired and congenital lower limb loss. *Journal Of Pediatric Orthopedics, Part B*, 20(3), 178-183. doi:10.1097/BPB.0b013e3283449362
- Varni, J. W., & Setoguchi, Y. (1992). Screening for behavioral and emotional problems in children and adolescents with congenital or acquired limb deficiencies. *American Journal of Diseases of Children (1960)*, 146(1), 103-107.
- Varni, J. W., & Setoguchi, Y. (1993). Effects of parental adjustment on the adaptation of children with congenital or acquired limb deficiencies. *Journal of Developmental and Behavioral Pediatrics*, 14(1), 13-20. doi:10.1097/00004703-199302000-00003
- Vasluian, E., de Jong, I. M., Janssen, W. M., Poelma, M. J., van Wijk, I., Reinders-Messelink, H. A., & van der Sluis, C. K. (2013). Opinions of youngsters with congenital below-elbow deficiency, and those of their parents and professionals concerning prosthetic use and rehabilitation treatment. *Plos ONE*, 8(6), 1-9. doi:10.1371/journal.pone.0067101
- Vasluian, E., van der Sluis, C. K., van Essen, A. J., Bergman, J. H., Dijkstra, P. U., Reinders-Messelink, H. A., & de Walle, H. K. (2013). Birth prevalence for congenital limb defects in the northern Netherlands: a 30-year population-based study. *BMC Musculoskeletal Disorders*, 14323. doi:10.1186/1471-2474-14-323
- Wallander, J. L., & Varni, J. W. (1992). Adjustment in children with chronic physical disorders; Programmatic research on a disability-stress-coping model. In A. M. LaGreca, L. Siegal, J. L. Wallander, & C. E. Walker (Eds.), *Stress and coping with pediatric conditions* (pp. 279-98). New York: Guilford Press.
- Wallander, J.L., & Varni, J.W. (1998). Effects of paediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry*, 39(1), 29-46. doi:10.1111/14697610.00302

- Weir, S., Ephraim, P., & Mackenzie, E. (2010). Effects of paediatric limb loss on healthcare utilisation, schooling and parental labour supply. *Disability and Rehabilitation*, 32(24), 2046-2055. doi:10.3109/09638288.2010.481028
- Yardley, L. (2000). Dilemmas in qualitative health research. *Psychology & Health*, 15(2), 215-228. doi:10.1080/08870440008400302
- Ylvén, R., Björck-Åkesson, E., & Granlund, M. (2006). Literature review of positive functioning in families with children with a disability. *Journal of Policy and Practice in Intellectual Disabilities*, 3, 253-270. doi:10.1111/j.1741-1130.2006.00089.x

Table 1.*Participant demographics*

Participant Pseudonym	Child's Age	Childs Gender	Years/Months prosthesis available	Location of Limb Difference	Nature of Limb Difference	Residing Country	Data Collection Method
Lisa	10	Female	10 years 2 months	Upper Limb	Congenital	UK	Face-to-face
Francis	7	Male	6 years 6 months	Upper Limb	Congenital	UK	Telephone
Jill	14	Female	13 years 3 months	Lower Limb	Congenital	USA	Telephone
Clare	9	Male	8 years	Lower Limb	Congenital	UK	Telephone
Ruth	8	Female	5 years	Lower Limb	Acquired	UK	Telephone
Elizabeth	11	Male	10 years 4 months	Lower Limb	Congenital	USA	Telephone
Patricia	11	Male	8 years 5 months	Lower Limb	Congenital	UK	Telephone

Table 2.*Final Themes and Individual Participant Themes*

Final theme	Participant Themes
<p>Theme 1: Managing the initial emotional experience through the development of coping resources</p>	<p>Lisa: Making sense of the initial shock: adjusting and adapting to a new idea of parenthood Lisa: Coping by becoming an advocate for child and predicting future challenges Francis: “Getting our head around everything”: making sense of the uncertainty and challenges of parenting a child with limb difference Clare: “A lot easier than I imagined”: initial expectations not matched the reality of parenting a child with limb difference Patricia: Coping through the strength of positivity and putting child’s needs first Patricia: Parental growth through learning and reflecting on experiences Jill: Managing the frustrations of prosthesis services: advocacy and staying connected Jill: The role of positive attributions in making sense of the experience of being a parent of a child with limb difference Ruth: Moving through feelings of regret and toward adaptation Ruth: Coping as a family through a focus on normality Elizabeth: Tackling the challenges through action Elizabeth: Providing the conditions to allow their child to thrive</p>
<p>Theme 2: Opportunities through prosthesis use and its relationship with ‘normality’</p>	<p>Lisa: A desire to keep artificial limb use options open to child Clare: Limb difference as irrelevant: prosthesis use enabling ‘normality’ Jill: Prosthesis use, ability and normality: instilling confidence and self-esteem in child Jill: Managing the frustrations of prosthesis services: advocacy and staying connected Ruth: Coping as a family through a focus on normality Elizabeth: Providing the conditions to allow their child to thrive Francis: Attitudes toward difference: critically considering benefits of prosthesis use Patricia: Pride in child’s ability to overcome challenges: a mirroring of Patricia’s own coping resources</p>

Theme 3: Managing and making sense of social reactions toward their child

Lisa: Negotiating the reaction of others toward child's limb difference

Clare: Social reactions and attitudes as the biggest challenge: a tension with acceptance

Ruth: Making sense of social reactions: hope around inclusivity

Elizabeth: Social responses as the biggest challenge: connecting with a sense of emotional pain

Patricia: Child as the same as others: the challenge of social reactions

Francis: "Getting our head around everything": making sense of the uncertainty and challenges of parenting a child with limb difference

Francis: Gaining support through charities and media visibility: the importance of positive support

Jill: The role of positive attributions in making sense of the experience of being a parent of a child with limb difference

Theme 4: The intrinsic role of support: developing a collective connection and enabling shared knowledge

Lisa: The importance of creating a support network and learning from each other

Lisa: A desire to keep artificial limb use options open to child

Francis: "Go away and figure it out yourselves": surprise and disappointment with healthcare support and prosthesis provision

Francis: Gaining support through charities and media visibility: the importance of positive support

Clare: Valuing prosthesis support and the prosthetist: feeling supported and lucky with care received

Jill: Strength through the support of others

Ruth: A discrepancy between practical and emotional aspects of healthcare support

Ruth: Placing a high regard and value on emotional support

Elizabeth: Gratitude and appreciation toward support

Patricia: The importance of a shared connection

Appendix A: Semi-Structured Interview

Demographic Information

Before commencing the interview could you briefly tell me how old your child is and the nature of their limb difference?

Parenting

What have been the key challenges or adaptations to parenting a child with limb difference?

How have you managed these challenges?

Have there been any benefits?

Has there been any impact to your family system? If so, what are some of the difficulties or positives encountered? Who, if anyone, in the family has been impacted?

How have you managed your own needs when parenting a child with limb difference?

Limb Difference and Prosthesis Use

What was your understanding of limb difference and prosthesis use before you had your child?

What does it mean to you now?

How did you decide for your child to be provided with an artificial limb?

Did you seek support in this decision-making process (family, friends, healthcare professionals)? If so what is your experience of receiving this?

How would you describe your relationship with the healthcare services supporting your child? How have you experienced this support?

Have there been any disagreements with healthcare professionals involved in your child's artificial limb use? If so, how have these been managed? If not, why do you think that is?

Prosthesis Use and Parenting

How has your child adapted to using an artificial limb?

Has this brought any challenges in your role as a parent?

How do you think your child feels about their artificial limb? What experiences have you had that has made you think this?

Have there been any benefits to your child in using an artificial limb?

Do you think your child has been impacted in anyway by having a limb difference? (school, friendships)? Has using an artificial limb changed this in anyway?

Final Thoughts

If you had to give advice for new parents of a child with limb difference what would it be?

If you could give advice or make any changes to the healthcare services involved during your journey of parenting a child with limb difference what would it be?

Specifically, to artificial limb use, what advice would you give to prosthesis services, to consider parents' experiences?

Thank you for taking part, do you have any final thoughts or reflections about your experiences of parenting a child with limb difference?

Appendix B: Initial Coding Excerpt

Initial Codes	Transcript
<p>“I think just encouraging them to know that are not different to anybody else” – Social stares from other children as a challenge to this</p> <p>Mum feels child is oblivious to stares but she isn’t</p> <p>Mum feels child is aware he isn’t different and can do things better than others</p> <p>Sense of parent attributing personal growth in child – “get stronger every time”</p> <p>Child had difficult social experience at school. Hard to hear for mum – mum made excuses for the child – ‘Stump’ example</p> <p>Mum upset at hearing about other child’s reaction, wanted to act but child stopped her</p>	<p>Just thinking about parenting, what do you consider to have been the key adaptations or challenges to parenting a child with limb difference?</p> <p>I think just encouraging them to know that are not different to anybody else. To make sure they know that as well, as well as myself. I think the biggest challenges are the staring and the other children who look and like to see what he has got which is different from everyone else.</p> <p>How have you managed that particular challenge?</p> <p>Quite well actually. I mean [child] has overcome stuff like that. He is now oblivious to other people looking at him so I think it’s probably more me that sees other people looking at him. But [child] became aware that he isn’t different from anybody else, and he can do things better than other people.</p> <p>How do you feel he has got to that point? Do you think that is anything you have instilled in terms of his parenting?</p> <p>He is just, he seems to be a strong kid, he has his own way of adapting and he just seems to get stronger every time.</p> <p>Has there been an example where that has happened?</p> <p>Well [child] just started high school in August, so one example is, he wasn’t wearing the limb he actually had his crutches, and there was a girl in the dinner queue who made a sick noise, when she saw his stump, and when [child] came home and told me, he wasn’t fazed by it at all. I was more annoyed by the fact that this had happened so I said ‘oh she probably hasn’t seen that before’, making excuses for the girl who had done this.</p> <p>How did that make you feel?</p> <p>I was a little upset by it I was actually going to phone the school and say to them to approach this girl and ask her why this had happened. And [child] was like ‘no its fine mum, its fine’.</p>

Appendix C: Summary Table of Individual Participant Themes

Participant	Themes
Lisa	<p>Theme 1. Making sense of the initial shock: adjusting and adapting to a new idea of parenthood</p> <p>Theme 2. Negotiating the reaction of others toward child's limb difference</p> <p>Theme 3. Coping by becoming an advocate for child and predicting future challenges</p> <p>Theme 4. A desire to keep artificial limb use options open to child</p> <p>Theme 5. The importance of creating a support network and learning from each other</p>
Francis	<p>Theme 1. "Getting our head around everything": making sense of the uncertainty and challenges of parenting a child with limb difference</p> <p>Theme 2. "Go away and figure it out yourselves": surprise and disappointment with healthcare support and prosthesis provision</p> <p>Theme 3. Attitudes toward difference: critically considering benefits of prosthesis use</p> <p>Theme 4. Gaining support through charities and media visibility: the importance of positive support</p>
Jill	<p>Theme 1. The role of positive attributions in making sense of the experience of being a parent of a child with limb difference</p> <p>Theme 2. Prosthesis use, ability and normality: instilling confidence and self-esteem in child</p> <p>Theme 3. Managing the frustrations of prosthesis services: advocacy and staying connected</p> <p>Theme 4. Strength through the support of others</p>
Clare	<p>Theme 1. "A lot easier than I imagined": initial expectations not matched the reality of parenting a child with limb difference</p> <p>Theme 2. Limb difference as irrelevant: prosthesis use enabling 'normality'</p> <p>Theme 3. Social reactions and attitudes as the biggest challenge: a tension with acceptance</p> <p>Theme 4. Valuing prosthesis support and the prosthetist: feeling supported and lucky with care received</p>
Ruth	<p>Theme 1. Moving through feelings of regret and toward adaptation</p> <p>Theme 2. Coping as a family through a focus on normality</p> <p>Theme 3. A discrepancy between practical and emotional aspects of healthcare support</p> <p>Theme 4. Placing a high regard and value on emotional support</p> <p>Theme 5. Making sense of social reactions: hope around inclusivity</p>
Elizabeth	<p>Theme 1. Tackling the challenges through action</p> <p>Theme 2. Providing the conditions to allow their child to thrive</p> <p>Theme 3. Social responses as the biggest challenge: connecting with</p>

	a sense of emotional pain Theme 4. Gratitude and appreciation toward support
Patricia	Theme 1. Coping through the strength of positivity and putting child's needs first Theme 2. Pride in child's ability to overcome challenges: a mirroring of Patricia's own coping resources Theme 3. Child as the same as others: the challenge of social reactions Theme 4. The importance of a shared connection Theme 5. Parental growth through learning and reflecting on experiences

Appendix D: Audit Trail Example ‘Lisa’*Theme 1. Making sense of the initial shock: adjusting and adapting to a new idea of parenthood*

Initial Codes	Narrative Summary	Participant Quotes
<ul style="list-style-type: none"> • Initial shock, diagnosis not picked up by the scan – “Shock to us all” • Lost idea of what having a child was, parent imagined them “riding a bike” • Having a child with limb difference adds an extra stress – “I was also trying to potty train her” • Tests to see if something “more serious” • Worry as consuming the initial experience of being a parent • Worrying about the arm had benefits in that she didn’t worry about things other parents might • Demands of multiple appointments – “lots of worry”- additional stresses on top of being a new parent • Would have liked to know diagnosis in advance to save “immense amount of shock” and to get “the reassurance” that the baby will be fine • Genetic counselling suggested birth of child with limb difference as “blip” • Parents feel they missed out on this reassurance due to diagnosis at birth • Parents didn’t find out about limb difference while pregnant, parent feels most parents do • Parent had no prior contact with limb difference while pregnant, not something that was in their thinking or life • Parent feels knowing before birth may have saved the end of pregnancy period as being “horrible” 	<p>This theme reflects the shock and subsequent impact to Lisa’s idea of parenthood after learning that her child was born with a limb difference. Lisa only found about her child’s limb difference at birth and this seems particularly relevant to how Lisa makes sense of the shock and worry that seems to permeate her early experiences of being a parent. Lisa talks about missing out on the reassurance she feels she would have been given if her child’s limb difference was diagnosed in pregnancy, indicating a lot of uncontained feelings after giving birth to her child.</p> <p>This was Lisa’s first child and there is a feeling of venturing into the unknown and having to adjust her preconceptions of parenthood. The initial stages were overwhelming for Lisa and there was a sense this meant she had to find extra resources to cope, above and beyond what she expected as a new mother. Lisa worried about how her child would cope with having a limb difference and there is a sense this changed expectation of being a mother of a child with limb difference was not easy for her</p>	<ul style="list-style-type: none"> • “Initially it was the shock, I didn’t know she was going to born without an arm, it hadn’t been picked up on the scan so it was a huge shock to us all when she was born” • “obviously you are pregnant you think about your child and imagine them doing things like, going to brownies or riding a bike or going to school and then obviously you suddenly think how is she going to do that” • “Initially she had a lot of tests because they didn’t know if it was just the arm or if it was something else more serious wrong with her” • “Just worrying really, worrying about the future how people would be with her and how she would manage and cope” • “but I think partly because I was worrying about the arm and the other things, how she was going to manage, rather than worrying about perhaps what new mums worry about, about I don’t know how many bottles they have had or nappy’s or that sort of thing, so I didn’t sort of have the brain space for that” • “Obviously early days, she was my first child, getting to grips with looking after a baby, but also a lot of worry” • “You know, if she had been born with two arms, our life would have been very different. I would

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|--|---|---|
| <ul style="list-style-type: none"> • New parents can get upset when meet a child with limb difference but “it’s quite useful” • Burden of appointments, referral that were not correct, feeling that doctors were unsure who to refer to. • Demands of appointments and having to tell your story over • Experience of always having to talk about child’s arm even with unrelated conditions such as a rash. • Parent able to rationalise early appointments burden “it did settle down” • Sheer number of appointments led to other health issues being picked up which parent feels wouldn’t have otherwise. • Early days “overshadowed” with health appointments • On reflection would want less referrals when child was born as this was an overwhelming aspect of adjusting to birth of child | <p>following the birth of her child.</p> <p>Lisa discusses the subsequent burden of appointments that occurred and very much something she wasn’t envisaging as a parent for the first time. There is a sense that by reflecting on these initial feelings, Lisa went through a process of adjusting to the demands of parenting a child with a limb difference and her experiences have not matched the levels of initial worry and stress she experienced. This comes through some of the language she uses, especially around referrals to subsequent appointments and tests searching for something “more serious”.</p> | <p>have certainly had a lot easier intro to parenthood”</p> <ul style="list-style-type: none"> • “at first they referred us to a lot of, they referred us to a plastics surgeon, I think they just sort of didn’t know where to refer us” • “I can sort of remember those early days being quite overshadowed with hospital appointments” • “certainly in the early days it was just obviously having to tell your story again and go over things again and relive things” • “It really just wasn’t on my radar at all. I can’t remember ever seeing anyone without a hand or an arm. I had certainly had no direct contact with amputees” • “Usually they see mums when you are still pregnant, they like to get you in the system as soon as you find out” • “Think again a lot of them (parents) get very upset, because it is upsetting to suddenly see a child when you have never seen one before, but it’s quite useful” • “The only thing I would say in the initial days would have been the referral, just referring us to the [limb centre] rather than referring us to plastics and various other people. • “Again I don’t think everybody does it but I would have liked to have known in advance but it would have saved the immense amount of shock” |
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Theme 2. Negotiating the reaction of others toward child's limb difference

Initial Codes	Narrative Summary	Participant Quotes
<ul style="list-style-type: none"> • Mum was worried about how other children would treat her daughter, will they accept her, or even over protect her – sense of it being a balance • Fears about social reactions – something I had to “get over” • Daughter can get frustrated if people ask more than once about arm – mum feels must be hard for her daughter to deal with stares “it adds another layer for her” • Experience with health visitor difficult, mum feels in hindsight she was “like a rabbit in the headlights” • Parents own mother is “obviously” fine with her • Daughter can get fed up with stares- mum feels she accepts it; she doesn't cry about it but can sometimes have a “moan” • Feels parents and children at primary school don't even see limb difference anymore, “it's not an issue at her primary school”- sense parent feels this is a good thing • Worrying about the impact of secondary school and transition • Parents family expressed shock at the news but knew family would be fine when informed of child's limb difference, didn't cross her mind that they wouldn't 	<p>This theme represents the central impact of societal views in the experience of parenting a child with limb difference. There is initially the impact this has on Lisa herself, including worrying about the possible reaction of close family members such as her mother. She reflects on this, and feels she knew they would accept her daughter and support her, but there is a sense it was a concern initially.</p> <p>The theme develops to include the reactions of the public and the impact on her daughter; there is a sense that Lisa feels a real need to want to protect her daughter. However, Lisa feels that the children and parents at her primary school no longer see her child's limb difference and that it isn't an issue. There is a sense that Lisa feels that her child's limb difference should always be treated this way but that it is not always the case. Transitioning to secondary school is a concern and a sense of not being able to control how this experience may be for her child.</p> <p>Integrated within societal views is the central aspect of wearing a prosthesis and Lisa recalling that one reasons her daughter likes to wear one is so people do not stare at her. Lisa also seems to value her daughters sense of humour regarding her prosthetic limb, which she feels can protect her child and serves a need to negotiate the potential negative reaction of others.</p>	<ul style="list-style-type: none"> • “I worried how the other children would be with her, I worried about how the staff would be with her, whether they would try and do too much for her, but at the same time I didn't want her to be overlooked or left behind” • “My mum is absolutely fine with her” • “She doesn't mind people asking once. But she gets frustrated, particularly little children” • “Whereas, I mean it must be daunting, wherever she goes she has that initial first few minutes of either looking or people asking her. I mean she does tell them I was born like that but it is obviously wherever she goes and any new experiences it is an extra layer for her” • “She is aware that people are going to be asking her and it's just getting over that initial asking her” • “I just worry really if the children, how they will take to her. Also I didn't want them babying her and trying to do things for her” • “Obviously going forwards, she will be mixing with a lot of different kids at her secondary school. She is at a small school now, she will be going to a much bigger school, a lot more children to mix with” • “She has willingly showed it off to her friends up the street. She put it on to show her friend up the street when she first got it and didn't tell her friend up the street, so were sat in her

-
- Worrying about how others will treat child, sense of balance between over-protection and not doing enough
 - Mum feels husband had worst job, telling people, sense that social reactions were a worry, other people's shock
 - Daughters reason for wanting a prosthesis so people will not stare
 - Child has a sense of humour about arm - surprised friend up street

bedroom and her friend hadn't even noticed and then [child] said 'ah haven't you even noticed what is different about me' and they were laughing about it. So she has definitely got that sense of humour with it as well"

- "No I knew they would be fine it was just that initial telling them and the shock but they have all accepted her and been absolutely fine with her, no that never crossed my mind I knew they would"
 - "telling people going out with her and people saying things or looking, just getting over that initial, whenever I went to baby group or anything like that it always something that had to be sort of got over, rather than just turning up like any other mum with a baby"
-

Theme 3. Coping by becoming an advocate for child and predicting future challenges

Initial Codes	Narrative Summary	Participant Quotes
<ul style="list-style-type: none"> • Time helped parent to adjust to challenges • Realisation that it is not a “massive obstacle” in life • Life totally different after having a child with limb difference but parent pragmatic “but that’s life, things develop” • Husband protective of daughter [mum laughing]. • Mother feels need to stand up for her daughter, advocate, be proactive and challenge perceived discrimination • Mum had to do things herself, precursor of own change, had to seek support herself • Proactive to seek support from charity • Some worries about initial prosthetic fitting but important “to sort of go with it” • Not chose to know about limb difference, it’s the “hand fate has dealt” • Mum feels she knows her child best and having to worry about things other parents would take for granted e.g. making a cake at school. Can’t just turn up “like the other kids” • Importance of having to think ahead, as others won’t do it for their child. Mum as the advocate • Feeling that parent’s needs are thought about by professionals but not able to see own coping resources as “you have no choice” but to cope • Good to know that professionals feel mum is 	<p>This theme reflects the journey toward adjusting to being a parent of a child with limb difference. Lisa discusses her experiences with a sense of reflection and pragmatism. She acknowledges her daughter does have unique challenges but reflects that her child’s limb difference has not been a big obstacle in their lives. She is now able to offer solace to other parents of children with limb difference who may have similar worries to those Lisa initially had. There is a sense of satisfaction that Lisa has been able to cope with the challenges she has been presented with.</p> <p>The theme does suggest Lisa has developed a number of coping resources to get to this appraisal that her daughters limb difference is not a big limitation. There is a sense of advocacy and planning, thinking ahead to potential difficulties. At the same time there is an acknowledgment that she had to rely on the expertise of her prosthetist and “sort of go with it”.</p> <p>Lisa seems to place value on her ability to seek out support and in effect be the precursor of change herself. There is a sense of the need for both herself and her husband to take a protective stance with their daughter to ensure she is treated fairly and given the same opportunities as her peers.</p>	<ul style="list-style-type: none"> • “I think, time has definitely helped and seeing that she is fine and that she can do things and it hasn’t been a massive obstacle in her life, she will go on to do whatever she wants to do and most of the time, it just doesn’t even come up, it isn’t an issue anymore” • “I mean my life is totally different to what it was 11 years ago, but that’s life, things develop” • “he doesn’t like any suggestion of anyone doing anything that might be, what shall I say, not treating her as she should be treated [laughs], or any suggestion of her being left out or anything like that” • “I do stand up for her in terms of probably how I approach things” • “As she has got older people do say to me, ‘oh you do cope very well’ and ‘aren’t you good’ and things like that, but obviously you do not know it’s just how you do it. You have no choice, but I have had that commented to me” • “Obviously it makes me think ‘well I must be doing something right’. But I also think well I’ve just done what was natural what, how I’ve thought I should do things” • “things like they were cooking lemon drizzle cake at the home ec lab and she’s going to probably need equipment, she’s going to need just a bit more thought and planning, rather than just turning up with all the other kids to

coping- validation that they are doing a good job

- Would tell other parents “it’s not the end of the world”
- Importance to communicate child limb difference in advance, not in a defensive way. Letting others know so issues less likely to arise. Mother putting self in others shoes
- “I’ve always tried to be one step ahead”

make the lemon drizzle cake, Bunsen burners in science labs things like that, again at the minute in primary school she’s absolutely fine and manages, but there are going to be some more challenges and thought processes coming up in the next few years

- “I’ve always tried to be one step ahead”
 - “I mean my life is totally different to what it was 11 years ago, but that’s life, things develop”
-

Theme 4. A desire to keep artificial limb use options open to child

Initial Codes	Narrative Summary	Participant Quotes
<ul style="list-style-type: none"> • Difficulty in training daughter to use myoelectric arm • Negotiating child wishes regarding prosthesis use with reassurance from limb centre, child goes through times of not wanting to wear one, but sense parent feels this is child's choice to make • Mother wanting to keep attending limb centre- wanting to keep options open to child • Making trips to the limb centre fun was important, not wanting them to be seen a negative light • Was told if they do not have one while young then child unlikely to choose to wear one as an adult • Cosmetic arm easier for child to put on, she paints nails and calls it a nickname • Challenges of managing a child with a myoelectric arm- she put it in sand at nursery • Child still able to use myoelectric "like riding a bike" • Importance of not forcing child to wear prosthesis so doesn't try- child knows best • Feels child accepts arm but chooses not to wear it as can get in way "she can do less with it on" • Still has same prosthetist- sense of trusting relationship important • Referred to prosthetist at birth- positive 	<p>This theme represents the challenges that occur with the integral feature of parenting a child with limb difference; the process of prosthetic rehabilitation. Here, Lisa discusses some of the challenges that occur when being a parent of a child who uses a prosthesis. She discusses the tension and uncertainty it can cause when her child chooses not to wear one and the reliance on healthcare professionals, in this case her daughter's prosthetist in supporting her through this uncertainty. Lisa reports that simply providing her with reassurance was valuable to her. There is a sense she places a lot of value on the support her prosthesis provided and the trusting relationship they have developed. Lisa seems to want to give her child the option of using myoelectric prosthesis and describes the process of her daughter adapting to one as being difficult but ultimately for the greater good. She reports having been told that once her daughter has learnt how to use one she will not forget.</p> <p>There is also a negotiation between parent and child around prosthesis use; with Lisa not wanting to force her child to use one, but similarly keeping up attendance at the limb centre just in case. There is a strong sense of wanting to keep all options open to her child to give her the choice as she gets older.</p> <p>Lisa recognises the challenges of funding and service provision and seems to manage this tension through</p>	<ul style="list-style-type: none"> • "I mean you physically couldn't force them to put it on if they didn't want to. I've always tried to encourage, particularly when she was younger to at least try it, but I mean if they want to pull it they will pull it off and that's that" • "So if there was a queue for children to get a limb, [child name] has always been top of the queue because we were very willing to go to appointments and willing to work with them" • "So she had a myo-arm while she was still at nursery or preschool, that was one of the hardest things I have ever done to train her to use it" • "She knew it wasn't supposed to go in sand or water and one day she put it in the sand in nursery, she was only about 3, and she washed it afterwards to get the sand off. That was beyond [location] fixing it" • "We always try to make the trip to the hospital, we do something else, as well usually, go to McDonalds or shopping afterwards or something... We always do something like that so it's not just the limb centre" • "it's probably that relationship the fact that they know you and you are not having to have that same conversation over and over again. People do tend to see the arm and think there is a lot more wrong with her"

evaluation of service received

- Importance of relationship with prosthetist and prosthetist taking an interest in child, importance of knowledge. Not wanting him to retire as parent values relationship
- Importance of being willing to support healthcare professionals working with individuals with limb difference -compliance will be rewarded
- Limitations of NHS provision can be frustrating but parent feels they push for prosthetics as much as can
- Parent feels engaging with healthcare services and volunteering has enabled them to be first in line for new services or prosthetics
- Importance to develop relationship with healthcare professionals and less need to talk about child arm when not relevant.
- Parents feel it is important to support doctors with their learning. Would volunteer as a teaching case. "we'll get a better service"

compliance, such as volunteering her child as a teaching case. Once more this allows options regarding prosthesis use to be kept open to her child, and there is a sense of doing all she can for her child by keeping all options open for when her child is older. She seems to place value on giving back to the healthcare system particularly by better educating healthcare professionals around issues pertinent to limb difference.

- "She still has the knack to do the myo, they told me it was like riding a bike, apparently once you have learnt the knack with your little muscles, you don't lose it"
 - "She is quite happy with it; I mean she wants to paint its nails and things"
 - "they have been very willing to push things as far as they can in terms of provision"
 - "I've always taken the view that if it wasn't too inconvenient to us I would rather doctors learnt about something. Then I thought 'well if someone else comes along after us hopefully they perhaps won't get referred or we'll get a better service"
 - "Yes, I mean he's a lovely man, he's very experienced, very calm, very reassuring. He's excellent at his job, I will be very sorry when, every time he goes I say, you're not retiring yet are you!"
-

Theme 5. The importance of creating a support network and learning from each other

Initial Codes	Narrative Summary	Participant Quotes
<ul style="list-style-type: none"> • Joining social media was great for parents, helped to educate them about limb difference and normalise many of the worries they had – instant support • Important to create support network such as joining charity, helps to normalise worries and concerns and opens up opportunities such as a limb loss camp. This camp had a positive impact on child • Use of social media helped to keep parent informed of what is available regarding prosthetic use • Importance of peer support helps to normalise experiences, normalise worries • Support as education tool, knowing about adaptations and what forms to get from GP – helps to “think a few steps ahead” • Social media important and enables positive aspect of parenting a child with limb difference to be seen clearer • Face-to-face support with other parents also an important aspect of being a parent with limb difference • Diagnosis not picked up till birth so feels missed out on parent support network • Feeling that child going to camp was a good thing for child- acknowledgement that this was hard for child as well • Vast majority of children at camp not wearing 	<p>The final themes concern Lisa’s appraisal of the importance of support in overcoming some of the challenges associated with parenting a child with limb difference who uses an artificial limb. There is a sense there can be an ‘unknowing’ aspect to being a parent and a need to educate yourself to be kept informed around issues relating to prosthesis use. The use of social media has allowed Lisa to connect to other parents and she has found this hugely valuable. Lisa described wanting to support other parents and enjoys being able to give back to the limb difference community and is closely involved with a national charity supporting children with upper limb loss. This support network also interacts with her perceived need to advocate for her child’s best interests, as she feels it allows parents to educate themselves so they can think ahead and be proactive in supporting their child.</p> <p>Finally, this support network led to an opportunity for her daughter to attend a camp with other children with limb difference. It came as a pleasant surprise to Lisa that many children at the camp did not wear a prosthesis, and so normalised some of the worries she had in relation to her own daughter’s prosthesis use. It seemed to provide validation too that she was doing the right thing in her role as a mother by not forcing her child to wear a prosthesis.</p>	<ul style="list-style-type: none"> • “Again though a lad I know through Reach; I knew he’s got one of the latest model ones from a different limb centre. Again I spoke to them about that and I know [name] was the first one funded for that type of limb at [location]. • “I’ve never disagreed with them, I have always, but they are aware I guess that I am quite, informed” • “I’d say it’s not the end of the world, life will be fine and join [charity] and its good for parents to speak or get advice from other parents” • “It’s good to see other older kids doing well and managing. Also for [child] she went on her first [charity] holiday this year. You have to be 10 to go, it’s one of those activity holidays... That has been a massive positive boast for her this summer. She is going again next year though [laughs], that’s what she has said! She went all the [location] all on her own, she didn’t know any of the other children going” • “I don’t think anyone had a prosthetic limb with them on that holiday. They were just as they were” • “But even more mundane stuff, you know like people put pictures you know of babies crawling and things, other mums say ‘oh I was worrying about that’ and then there is a picture

a prosthetic arm

of some little baby”

- “The face-to-face meet-ups with [charity] are very good because again, people will speak to you, you can speak to people, and just to see, other kids running round”
-

Appendix E: *Psychology and Health* Instructions for authors

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Section Three: Critical Appraisal

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Abstract

The experience of completing this thesis has led to the development of research skills and provided theory-to-practice links relevant to my clinical work. Inevitably, the process of conducting research has provided the opportunity to overcome several barriers and to think about professional issues relevant to clinical psychology as a profession. In this paper, I will look to offer a critical reflection of the empirical paper and have structured the paper into two parts. Part 1: 'The Role of *'me'* in the Research' documents personal reflections regarding my relationship with the topic area, and the process of navigating methodological issues to ensure fidelity with the principles of interpretative phenomenological analysis (IPA). Part 2: 'Toward Qualified Life' considers my relationship with the thesis as it draws to its conclusion, and offers a consideration of the clinical implications pertinent to the next stage of my career.

Part 1: The Role of 'me' in the Research

Throughout this thesis journey it has been important to consider my own thoughts and feelings regarding the research. Finlay (2002) suggests our behaviour as researchers will affect participant responses and so has the potential to influence and shape research findings. Tufford and Newman (2012) suggest the narrative of objectivity within qualitative research has dissolved. Thus, there appears to be an increased awareness, acceptance and embracement of the subjectivity inherent within qualitative research through the process of reflexivity. Etherington (2004) suggests this involves an awareness of our personal responses, so we can make informed choices relating to how we use them. It includes an awareness of the personal, social and cultural contexts in which we live. In research settings, this involves the researcher developing an understanding of their own active role throughout the research process (Willig, 2010). The importance of self-awareness within phenomenological research is vital, with a central tenet the bracketing of any presuppositions (Ashworth, 1996). This can relate to the influence of previously conducted research, as well as the researcher's own personal beliefs related to the research area of interest. As such the use of a reflexive diary was a key aspect to developing a sense of self-awareness throughout the research process. I will now document this process of reflexivity to illustrate my relationship with the research topic, and how I navigated methodological issues to ensure fidelity with the principles of IPA.

My Relationship with the Research Topic

On the Lancaster DClInPsy programme we are afforded autonomy to pursue our own research interests. I felt it was important to consider my clinical interests, as I firmly believe in the value of research influencing clinical practice. A benefit of clinical psychology being a doctoral programme is that we are trained to both critical evaluate, and contribute, to the

evidence base to inform practice (British Psychological Society, 2014a). Throughout training I have developed a clinical interest in paediatric psychology and so wanted to pursue a thesis that would be related. Through discussions with my research supervisor we were able to collaboratively agree on a research topic that had a fit with both of our research interests.

The British Psychological Society's (2014b) *Code of Human Research Ethics* suggest research within the discipline should acknowledge a social responsibility regarding research outcomes: "the aim of generating psychological knowledge should be to support beneficial outcomes" (p.10). At this stage I was keen to ensure my research idea was serving a clinical need. Through researching the literature, it was clear the research would fill an apparent gap. In addition, I contacted a local charity supporting parents of children with limb difference, who confirmed the under-representation of parents' voices and the need for research that could illuminate possible support needs.

I was drawn to the exploratory nature of the research and a chance to prioritise the experiences of an under-represented population. Admittedly, I did experience anxieties related to researching a topic area I knew little about. I had neither personal nor clinical experience relating to limb difference or prosthesis use. I was also keen to ensure the applicability to clinical psychology: the involvement of an experienced clinical psychologist who formerly worked in paediatric settings helped to allay this anxiety.

I was specifically attracted to using IPA given its focus on prioritising lived experience and sense-making. IPA is considered to fit within a realist epistemological framework which assumes language can be used to access the inner experience of participants (Crossley, 2000). IPA also acknowledges that achieving an understanding of participant experience is complicated by the active role of the researcher and the dynamic social process (Smith & Osborn, 2008). IPA therefore seemed a good fit with my own

epistemological position where I consider myself a critical realist (Maxwell, 2012): that is, I subscribe to an ontological realism, the idea that there exists an external 'truth' independent from our perceptions and constructions, but that construction of truth is always influenced by the social context with which it exists.

Thought was given to the experience of interviewing parents and issues relating to disability. I am not a parent, nor do I have any family or friends who have experienced limb difference. I wondered how this may impact the interview and my ability to stay present with the parents' experiences and understand issues from their perspective. After conducting the first interview, I was struck by its emotive nature, and felt a sense of humbleness regarding the bravery and resilience Lisa had shown. An extract from my reflexive journal after the interview read:

Struck by the privileged position we find ourselves in – hearing participant's intimate stories. Surprised by the emotive nature of interview. Being respectful of this felt important. (13/09/2016)

Similarly, participant experiences related to issues around disability, and their own definitions of disability were hard to navigate, and provided the opportunity for reflection and learning. For instance, Clare discussed that she did not consider her child to have a disability because of the lack of limitations it has on her child's life. I remember this striking me as a powerful statement at the time of the interview, and left me to consider my own definition of disability and views around diversity and inclusivity. Fawcett and Hearn (2004) discuss some of the issues of researching 'others'. They advocate for researchers to take a reflexive stance relating to a critical engagement of both the social bases of the researchers acquired knowledge and an understanding of the relevant political agendas. In this instance, I was able

to become aware of personal 'blind spots' in relation to my knowledge and understanding of issues that were of importance to participants.

The Navigation of Methodological Considerations

IPA is predicated on the philosophical approaches of phenomenology, idiography and hermeneutics (Smith, Flowers & Larkin, 2009). Phenomenology is concerned with the central experience of participants, using their own language as vehicles to access their perspectives and the meaning ascribed to their experiences. Related to this, idiography is concerned with the process of prioritising the uniqueness of the individual and that each participant's experiences are unique to them. Hermeneutics is concerned with interpretation: IPA acknowledges that understandings of participant experiences are complicated by the researcher's own preconceptions. As such, "the researcher is trying to make sense of the participant trying to make sense of their world" (Smith & Osborn, 2008, p.53); referred to as a *double hermeneutic*. The philosophical assumptions of IPA detailed above, need to be considered in the sampling, the collection of data and subsequent analysis. I will now describe some of the challenges this presented.

Homogeneity. Samples are selectively chosen on the assumption that they allow access to a particular perspective in relation to the aims of the study (Smith et al., 2009). In IPA studies, homogeneity of the sample is a fundamental aspect and refers to the way in which the sample shares key characteristics. Murray and Wilde (in press) suggest participants within an IPA study are homogenous if they are bound by a particular experience of a phenomenon. In this way, homogeneity goes beyond demographic or 'measurable' characteristics of the sample. Of relevance to my study was a consideration of the parameters by which the sample could be considered to have a shared experience: this posed somewhat of a challenge. Given the research was exploratory in nature, key decisions in relation to the

sample were the nature (acquired versus congenital) and location (upper versus lower) of the child's limb difference and the child's age range.

When considering the nature of limb difference, I decided to include parents of children with both congenital and acquired limb difference. This decision was made on the basis that the research was exploratory and centrally concerned with prosthesis use. There was also a pragmatic element to this decision relating to recruitment. As I was recruiting through the social media accounts of relevant limb difference charities, I was unsure how successful this approach would be; therefore, I felt it important not to overly restrict the sample. Similarly, consideration of the type of limb difference, upper versus lower, was significant. It was important to consider whether parents' experiences would be fundamentally different based on the type of limb difference their child had experienced. The results of the study show that the inclusion of both upper and lower limb difference highlighted a significant divergence around the experience and meaning related to their child's use of a prosthesis concerning functionality. This would have been missed if I had chosen to focus exclusively on upper or lower limb difference. This seems to relate to homogeneity being an interpretative issue, and thinking carefully about how much variation can be tolerated within the focus of the study (Smith et al., 2009). I would assert that the decision to include parents of children with both upper and lower, congenital and acquired limb difference, enhanced interpretation relating to participant experiences and meanings.

The age range of the parents' children was another consideration. Having a field supervisor who had worked within paediatric psychological services was advantageous in thinking carefully about this and drawing on their clinical experience. As the study wanted to explore parents' experiences related to the prosthetic rehabilitation process, as well as experiences related to parenting a child with limb difference more generally, it was decided to focus on an age range that enabled the opportunity to best access these aspects of

experience. The age range of 5-16 was decided upon as it felt it best allowed the possibility to capture parental experiences across key developmental milestones and important life events. For example, the child able to verbalise thoughts and feelings, having begun attending school, and the transition to high school. In the sample recruited, the child's ages ranged from 7-14, however there was just one adolescent. Further research could look to address this, with adolescence often bringing a number of challenges for individuals with a visible difference (Rumsey & Harcourt, 2007).

Sample size. Smith et al. (2009) make an interesting point in relation to an almost implicit defensiveness in qualitative research regarding sample size that can perpetuate the use of larger sample sizes to deflect criticism from quantitative researchers. However, given the primary concern of IPA is a detailed account of individual experience, I was concerned that a large sample size could dilute the richness of analysis. This could have led to a weak level of interpretation around the convergences and divergences across participant accounts. Therefore, as Smith et al. (2009) suggest, the issue of sample size is about "quality, not quantity, and given the complexity of most human phenomena, IPA studies usually benefit from a concentrated focus on a small number of cases" (p.51). They do offer a rough approximation of a suitable sample size for professional doctorate research of between 4-10 participants. Given the guidance, a sample size of between 4-12 participants was decided in advance.

Data collection: The use of a semi-structured interview is a key method for data collection within IPA studies. Reflecting on its use, one of the areas of challenge concerned managing the tension between the role of the being a researcher as opposed to a trainee clinical psychologist. In my clinical work, I am trained to work collaboratively and to think carefully about the therapeutic relationship, which is formed over several sessions. In the researcher role, I only spoke to the participant once, with the content very much guided by

the research aims. I found many of the parent accounts emotionally moving and wanted to explore the emotional impact to support them through it. For example, Patricia discussed many strong emotions relating to being a young single mum and her belief that she could not cope in the role of a parent. I was aware I wanted to develop a collaborative understanding around her experiences that may have contributed to this belief. The use of supervision helped to explore this tension, and I feel this helped me reach a position where I could stay present with the participants' emotions, without a need to develop a joint understanding around the 'why'. This helped me to stay grounded with the research aims.

The current study utilised mediated communication methods which had the benefit of accessing participants from around the world. One parent was interviewed face-to-face and six over the telephone; two telephone interviews were conducted with parents residing in the USA. The decision regarding the interview format was based on a combination of participant preference and logistical considerations regarding locality.

This was the first time I had used telephone interviews; as such I experienced initial anxieties relating to the development of rapport. In response to this anxiety, it felt important to use my active listening skills to help support rapport and engagement. This involved the use of summarising and reflecting back participant responses to show the participant I heard them. This seemed to allow a deeper exploration of participant experience. Holt (2010) suggests while telephone interviews can lose some of the subtleties of non-verbal communication, they can have the benefit of allowing the researcher to stay grounded in the data and avoid the imposition of contextual information. Similarly, Novick (2008) discusses what they refer to as a bias against telephone interviews, with the common assumption that its use can compromise rapport and the quality of data produced. Novick suggests that there is limited evidence to match this assumption, and its use may promote the participant to feel relaxed and disclose sensitive information. I must admit I also had similar assumptions and

doubts concerning the use of telephone interviews, however my experiences are in keeping with the suggestions of both Holt (2010) and Novick (2008). Interestingly, Ward (2015) used grounded theory to explore participant views of taking part in telephone interviews.

Participants reported it to be a positive experience, with the benefits including the development of rapport and not feeling inhibited or judged.

A further anxiety in relation to the use of telephone interviews was managing any risk or safeguarding issues. Seeking support from both my research and field supervisor was a key element in navigating this issue. As such I ensured I had the addresses of all participants before commencing the interview. I also informed all participants of my duty of care to act on any risk issues. Although none arose, I planned to use my clinical skills to explore any risk or safeguarding issues, and would have sought support from both research supervisors to consider the effective management of the issue.

Analysis. A key aspect in IPA research is the bracketing of any assumptions or taken for granted truths that may influence data collection and the subsequent analysis (Murray & Wilde, in press). To look to become aware of and acknowledge any assumptions, I used a reflexive diary to note my thoughts and feelings both before and after each interview. This process proved helpful, as after the first interview, I became aware of the influence of previously conducted research regarding unsatisfactory healthcare experiences for adolescents with type-1 diabetes. An extract from my reflexive diary read:

Being aware of own assumptions- assumption around healthcare professionals perhaps not being helpful and the communication being didactic (13/09/2016)

This had the potential to influence the line of questioning and move experiences away from participants' own meanings. Therefore, being able to identify and bracket such assumptions safeguarded the potential influence on the analysis.

Part 2: Toward Qualified Life

Acknowledging the End

The production of the research paper has involved many months' work, from the initial planning stage and development of a research protocol, to the recruitment and write-up. Similarly, the systematic literature review has presented challenges, such as finding a novel topic of interest and navigating some of the inherent methodological challenges that are central to a meta-ethnography (Noblit & Hare, 1988). The process therefore has been academically demanding, emotionally challenging but also uplifting and rewarding.

I feel a mixture of emotions regarding this thesis process approaching the end. Part of my development throughout clinical training has been to become aware of my own emotional responses in relation to clinical work. Given the range of emotions experienced during the production of this thesis, which have ranged from frustration to joy, it feels important to acknowledge and reflect on my feelings regarding the thesis coming to an end. The importance of 'endings' in therapy are well written (e.g. Smith & Garforth, 2012) and associated with a range of powerful emotions for both client and therapist. Facilitating the expression of these emotions is an important part of the ending process. Writing this critical appraisal, I am struck by a feeling of excitement regarding the increase in personal time I will be afforded on its completion, and pride in relation to the work completed. I also feel a sense of responsibility to the participants to try to publish the research so their voices are heard. This is something which feels very important and a way of acknowledging and showing gratitude to the time and experiences the participants' generously shared. Additionally, I feel strongly that healthcare services should be informed by service user experience.

Moving Forward

As I move forward to the next stage of my career, it feels appropriate to explore the clinical implications of the research and how it can help shape both my professional development and clinical practice. The research has reaffirmed my views regarding the importance of exploring parents' support needs and enabled due consideration of issues related to the promotion of psychological well-being. This has left me to reflect on a quote which encompasses my motivation to work within paediatric psychological settings: "ordinary children facing extraordinary challenges" (Houghton, 2005, p.114); I feel this statement could extend to parents also.

The results of both thesis papers found parents described a range of powerful emotions when they were informed of their child's condition. This led me to reflect on a poem by Emily Perl Kingsley (1987) entitled "Welcome to Holland". For me, it beautifully encapsulates a parent's journey toward adjusting to a new understanding of being a parent. It personally inspires a feeling of warmth and optimism as she describes the process of beginning to appreciate the wonder of the unexpected destination of Holland, while acknowledging the loss related to the change in destination.

Both thesis papers discussed the importance of supporting parents own emotional well-being. The children's National Service Framework standard for hospital services (Department of Health, 2003), states that the mental health of young people and their families should be an integral component of child healthcare services. Interestingly however, there appears to be a lack of parity and equity around the distribution of psychological support within paediatric healthcare services across the UK (Mercer et al., 2015).

Within services supporting individuals with limb difference some services do have access to a clinical psychologist. They can work closely with the multi-disciplinary team (MDT) to provide psychological support (e.g. most locally to Lancaster at Aintree University

Hospital NHS Foundation Trust). The skills and knowledge clinical psychologists can bring to a medical MDT facilitates access to specialist psychological intervention. Furthermore, it allows system wide acknowledgment of issues related to psychological well-being and enables families to seek support in a non-stigmatising setting (Mercer et al., 2015). I feel increasing the awareness of issues related to psychological well-being, including the role of social factors, within a medical MDT can help shift prevailing biomedical cultures toward psychosocial perspectives. Within the guidelines for the physiotherapy management of adults with a lower limb prosthesis (British Association of Chartered Physiotherapists in Amputee Rehabilitation, 2012), there is some acknowledgement of a need to consider a patient's psychological well-being after they have been discharged; however it can read as almost an afterthought. This I feel needs to be addressed, with greater professional cohesion needed to bridge the gap between physical and psychological care. Having recently secured a role within a paediatric psychology service, I feel well-placed to continue to engage in this issue in the next stage of my career.

This thesis has also allowed a critical engagement with issues pertinent to both clinical psychology and disability research. Through the research I have become aware of the social model of disability. The model suggests the implicit social structures within society impose disability on top of an individual's impairment (Shakespeare, 1993), for example a building not being accessible to wheelchair users. Lankhorst et al.'s (2016) research could also be seen to provide a recent example of this amongst individuals with upper limb difference. They found older adolescents experienced difficulties finding employment, even though the participants themselves felt they were physically capable of completing the advertised role. This model fits well with my own views around the role and impact of social structures on psychological well-being.

The Division of Clinical Psychology (2011) encourages psychologists to consider the role of social and contextual factors when collaboratively developing psychological formulations with service users. However, Supple (2005) suggests psychological research can often be accused of perpetuating an assumption that disability is a physical problem, largely affected by an individual's thoughts and behaviours, with little acknowledgement of social or contextual factors. Simpson and Thomas (2015) explored a disciplinary divide between clinical psychology and disability studies, and suggest clinical psychology needs to look to engage more at societal and political levels to influence change. Moreover, Supple (2005) suggests psychologists need to reflect on their own assumptions regarding disability and evaluate how their training programme engages in the topic of disability. This has led me to reflect on the importance of clinical psychology programmes providing teaching related to social issues and barriers that may impact those with a disability. For me personally, I feel this research and engagement with some of these issues, has left me feeling more confident to work with and support children and their families with a physical impairment or difference, and to understand some of the systemic barriers contributing to their current difficulties.

Finally, the process of completing the research has led to due consideration of future research opportunities which can look to bridge the gap between disability and psychological research. I feel IPA, and its focus on meaning and sense-making, is well placed to provide a holistic understanding of the experience of disability and how it may relate to issues of relevance to clinical psychology. This may inform future research, for example exploring the meanings attached to using a prosthesis amongst children and adolescents, which may compliment and/or extend the findings from my research paper. Research could also further explore parents' experiences related to being informed about their child's limb difference, which could influence effective healthcare support during this challenging time.

Conclusion

It is hoped this critical appraisal has illustrated some of the pertinent issues regarding the development of the research paper, as well as illuminating my own role in shaping the research and ultimately the findings. This expression of reflexivity is a central part of IPA and allows the reader to critically consider the validity of the research (Yardley, 2008). I have also highlighted my reflections regarding the thesis process coming to an end, and discussed some of the clinical implications that I feel will be particularly relevant to the next stage in my career.

References

- Ashworth, P. (1996). Presuppose nothing! The suspension of assumptions in phenomenological psychological methodology. *Journal of Phenomenological Psychology*, 27(1), 1-25. doi:10.1163/156916296X00014
- British Association of Chartered Physiotherapists in Amputee Rehabilitation. (2012). *Evidence based clinical guidelines for the physiotherapy management of adults with lower limb prostheses*. London: The Chartered Society of Physiotherapy. Retrieved from www.csp.org.uk/sites/files/csp/secure/bacpar_amputee_rehab_guidelines_2012_1.pdf
- British Psychological Society. (2014a). Standards for doctoral programmes in Clinical Psychology (2014). Retrieved from http://www.bps.org.uk/system/files/Public%20files/PaCT/dclinpsy_standards_approved_may_2014.pdf
- British Psychological Society. (2014b). *Code of research ethics*. Retrieved from <https://beta.bps.org.uk/files/code-human-research-ethics-2014pdf>
- Crossley, M. (2000). *Introducing narrative psychology: Self, trauma and the construction of meaning*. Buckingham: Open University Press.
- Department of Health. (2003). *Getting the right start: National service framework for children standard for hospital service*. London: The Stationary Office.
- Division of Clinical Psychology. (2011). *Good practice guidelines on the use of psychological formulation*. Leicester: British Psychological Society.

- Etherington, S. (2004). *Becoming a reflexive researcher: Using ourselves in research*. London: Jessica Kingsley
- Fawcett, B., & Hearn, J. (2004). Researching others: Epistemology, experience, standpoints and participation. *International Journal of Social Research Methodology: Theory & Practice*, 7(3), 201-218. doi:10.1080/13645570210163989
- Finlay, L. (2002). 'Outing' the researcher: The provenance, process and practice of reflexivity. *Qualitative Health Research*, 12(4), 531-545.
doi:10.1177/104973202129120052
- Holt, A. (2010). Using telephones for narrative interviewing: A research note. *Qualitative Research*, 10, 113-121. doi:10.1177/1468794109348686
- Houghton, J. M. (2005). Paediatric psychology in the twenty-first century: Forward together? *Clinical Child Psychology & Psychiatry*, 10(1), 112-117.
doi:10.1177/1359104505048797
- Kingsley, E.P. (1987). *Welcome to Holland*. Retrieved 1st May 2017, from: <http://www.our-kids.org/Archives/Holland.html>
- Lankhorst, I. F., Baars, E. T., Wijk, I. v., Janssen, W. M., Poelma, M. J., & van der Sluis, C. K. (2016). Living with transversal upper limb reduction deficiency: limitations experienced by young adults during their transition to adulthood. *Disability And Rehabilitation*, 1-8. doi:10.1080/09638288.2016.1206632
- Maxwell, J. A. (2012). *A realist approach for qualitative research*. London: Sage.

- Mercer, A., O'Curry, S., Donnan, J., Stedmon, J., Reed, J., & Griggs, H. (2015). Delivering psychological services for children and young people with physical health needs and their families. *Child & Family Clinical Psychology Review*, 3, 71-81.
- Murray, C., & Wilde, D. J. (in press). Phenomenology and interpretative phenomenological analysis. In S. Cartwright, C. Walshe, & S. Brearley (Eds.), *Handbook of theory and methods in applied health research*. Cheltenham: Edward Elgar.
- Noblit, G. W. & Hare, R. D. (1988). *Meta-Ethnography: Synthesizing qualitative studies*. London: Sage.
- Novick, G. (2008). Is there a bias against telephone interviews in qualitative research? *Research in Nursing & Health*, 31(4), 391-398. doi:10.1002/nur.20259
- Rumsey, N., & Harcourt, D. (2007). Visible difference amongst children and adolescents: Issues and interventions. *Developmental Neurorehabilitation*, 10, 113-123. doi:10.1080/13638490701217396
- Shakespeare, T. (1993). Disabled People's Self-organisation: a new social movement? *Disability, Handicap & Society*, 8(3), 249-264. Retrieved from <http://www.tandfonline.com/toc/cdso19/current>
- Simpson, J., & Thomas, C. (2015). Clinical psychology and disability studies: Bridging the disciplinary divide on mental health and disability. *Disability and Rehabilitation: An International, Multidisciplinary Journal*, 37(14), 1299-1304. doi:10.3109/09638288.2014.961656
- Smith, A., & Garforth, K. (2012). Psychodynamic therapy. In S. Weatherhead & G. Flaherty-Jones (Eds.), *The pocket guide to therapy. A 'how to' of the core models*. (pp.77-100). London: Sage.

- Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative phenomenological analysis: Theory, method and research*. London: Sage.
- Smith, J. A., & Osborn, M. (2008). Interpretative phenomenological analysis. In S. A. Smith (Eds.), *Qualitative psychology: A practical guide to methods* (pp. 53-80). London: Sage.
- Supple, S. (2005). Getting our house in order. *The Psychologist*, 18(7), 418-420. Retrieved from <https://thepsychologist.bps.org.uk/volume-18/edition-7/disability-special-issue>
- Tufford, L., & Newman, P. (2012). Bracketing in Qualitative Research. *Qualitative Social Work*, 11(1), 80-96. doi:10.1177/1473325010368316
- Ward, K. (2015). Participants' views of telephone interviews within a grounded theory study. *Journal of Advanced Nursing*, 71(12), 2775-2785. doi:10.1111/jan.12748
- Willig, C. (2010). *Introducing Qualitative Research in Psychology: Adventures in theory and method* (2nd edition). New York: Open University Press.
- Yardley, L. (2008). Demonstrating validity in qualitative psychology. In J. A. Smith (Eds.), *Qualitative psychology: A practical guide to research methods* (pp. 235-251). London: Sage.

Section Four: Ethics Section

**Exploring the Experiences of Being a Parent of a Child with Limb Difference who has Been
Provided with an Artificial limb**

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Doctorate in Clinical Psychology

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**Faculty of Health and Medicine Research Ethics Committee (FHMREC)
Lancaster University**

**Application for Ethical Approval for Research involving
direct contact with human participants**

Instructions [*for additional advice on completing this form, hover PC mouse over 'guidance'*]

1. Apply to the committee by submitting:
 - a. A **hard copy** of the University's **Stage 1 Self Assessment (part A only)** and **Project Questionnaire**. These are available on the Research Support Office website: [LU Ethics](#)
 - b. The completed application **FHMREC form**
 - c. Your full research proposal (background, literature review, methodology/methods, ethical considerations)
 - d. All accompanying research materials such as, but not limited to,
 - 1) Advertising materials (posters, e-mails)
 - 2) Letters/emails of invitation to participate
 - 3) Participant information sheets
 - 4) Consent forms
 - 5) Questionnaires, surveys, demographic sheets
 - 6) Interview schedules, interview question guides, focus group scripts
 - 7) Debriefing sheets, resource lists

Please note that you DO NOT need to submit pre-existing handbooks or measures which support your work, but which cannot be amended following ethical review. These should simply be referred to in your application form.

2. Submit the FHMREC form and all materials listed under (d) by email as a **SINGLE attachment in PDF format** by the **deadline date**. **Before converting to PDF ensure all comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line*.**
3. Submit one collated and signed paper copy of the full application materials in time for the FHMREC meeting. If the applicant is a student, the paper copy of the application form must be signed by the Academic Supervisor.
4. Committee meeting dates and application submission dates are listed on the [FHMREC website](#). Applications must be submitted by the deadline date, to:

Dr Diane Hopkins
B14, Furness College
Lancaster University,
LA1 4YG
d.hopkins@lancaster.ac.uk
5. Prior to the FHMREC meeting you may be contacted by the lead reviewer for further clarification of your application.
6. Attend the committee meeting on the day that the application is considered, if required to do so.

1. **Title of Project:** Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.
2. **Name of applicant/researcher:** Mr James Oliver

3. Type of study

Includes *direct* involvement by human subjects.

Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. Please complete the University Stage 1 Self Assessment part B. This is available on the Research Support Office website: [LU Ethics](#). Submit this, along with all project documentation, to Diane Hopkins.

4. If this is a student project, please indicate what type of project by marking the relevant box/deleting as appropriate: (please note that UG and taught PG projects should complete **FHMREC form UG-tPG**, following the

procedures set out on the [FHMREC website](#)

PG Diploma Masters dissertation PhD Thesis PhD Pall. Care
 PhD Pub. Health PhD Org. Health & Well Being PhD Mental Health MD
 DClinPsy SRP [if SRP Service Evaluation, please also indicate here:] **DClinPsy Thesis**

Applicant Information

5. Appointment/position held by applicant and Division within FHM

Trainee Clinical Psychologist

6. Contact information for applicant:

E-mail: j.oliver1@lancaster.ac.uk

Telephone: [REDACTED] (please give a number on which you

can be contacted at short notice)

Address: Clinical Psychology, Div. Of Health Research,
Lancaster University, Lancaster, LA1 4YG

7. Project supervisor(s), if different from applicant:

Dr Craig Murray

8. Appointment held by supervisor(s) and institution(s) where based (if applicable):

Senior Lecturer, Lancaster University DClinPsy.

Email: c.murray@lancaster.ac.uk

9. Names and appointments of all members of the research team (including degree where applicable)

Dr Clare Dixon, Clinical Tutor, Lancaster University DClinPsy.

Email: c.dixon3@lancaster.ac.uk

The Project

NOTE: In addition to completing this form you must submit a detailed research protocol and all supporting materials.

10. Summary of research protocol in lay terms (indicative maximum length 150 words):

Those with limb difference have been shown to have an increased prevalence of emotional and behavioural issues and can have lower social competence than healthy peers (Varni & Setoguchi, 1992). Parenting a child with a disability has been shown to be related to increased parenting stress (Baker-Ericzen, Brookman-Frazee, & Stahmner, 2005). Ylven, Bjorck-Akesson and Granlund, (2006) suggest finding positive meaning in a child's disability can enhance coping and lead to a reduction in feelings of stress. To date, there is no research literature relating to parents' lived experiences of parenting a child with limb difference and how they negotiate the prosthetic rehabilitation process; this is an aspect unique to this population and a key element in parenting a child with limb difference. Smith and Campbell (2009) suggest a number of key issues for parents regarding the use of a prosthesis for their child. As such, the study is looking to provide insight into the lived experiences of parents, and give an understanding to some of the challenges that need to be navigated. The study is looking to address a gap in the literature by interviewing parents of children with limb difference who have been provided with an artificial limb

11. Anticipated project dates (month and year only)

Start date: August 2016

End date: May 2017

12. Please describe the sample of participants to be studied (including maximum & minimum number, age, gender):

The study is looking to recruit 4-12 participants. The minimum number of participants to be recruited will be 4. The sample will be international. Participants will be parents of children with limb difference who have been provided with a prosthesis. There are specific inclusion and exclusion criteria:

Inclusion:

- Parents to have a child with limb difference and have available to use an artificial limb
- Age range of child to be between 5-16 years old (primary and secondary school)
- Child's limb difference can be congenital or acquired through any event e.g. trauma or resultant from a health condition such as meningitis
- Child to have had the availability of a prosthesis for a minimum of 6 months
- Parents to speak English

Note on Inclusion:

- Rationale for this age range is that I want to capture parental experiences across key developmental milestones and important life events (e.g. having begun attending school, transition to high school, developing identity through puberty). The idea is to keep the age range broad because the research is novel and exploratory.
- In the event the study is oversubscribed, I will use a quota to choose particular participants which would provide the study with variety such as particular age ranges of children e.g. x amount of 5-7 year olds, x amount of 7-10 etc.
- If the study is not oversubscribed then I will include all participants who meet the inclusion criteria until the recruitment target is achieved.

Exclusion:

- Parents of children with a co-morbid health condition which may limit or preclude prosthesis use or any other physical and/or intellectual disability.

The rationale for this, is that the presence of any of these factors may mean parents are facing different challenges and experiences to those posed specifically by having a child with limb difference, who have been provided with an artificial limb.

13. How will participants be recruited and from where? Be as specific as possible.

Participants will be recruited via online charities. Recruitment will take the form of a three-point plan where possible.

1. Via relevant and amiable charities advertising and disseminating the participant information sheet via social media, home webpages and mailing lists

Relevant charities will be contacted via phone and/or email (see appendix D for draft email correspondence). Charities will be emailed the participant information sheet (appendix A) and the social media advertisement with an electronic link to the participant information sheet (appendix E), and asked to share this via their home webpage, Twitter and Facebook accounts. Furthermore, charities will also be asked if they are able to send the participant information sheet to their mailing lists. Finally, charities will also be asked to advertise the study in any publications they have and then sent an advertisement if they are happy to do so (appendix F).

2. Via Twitter

The lead researcher proposes to set up a research twitter account that can have a link to the participant information sheet (on a research advertising space for DCLinPsy students). The twitter account would be solely used to advertise the proposed study and to share the participant information sheet. Active online charities will be contacted and asked to consider 're-tweeting' my post (see appendix E), so other followers of that site can gain voluntary access to the study information, and email the lead researcher to take part.

3. Via Facebook

Active charities will be asked to share the participant information sheet on their Facebook page. No specific Facebook account will be set up by the lead researcher, rather the participant information sheet will be sent to charities and they will be asked to share it on their Facebook page. Again, participants can access the study details and get in touch with the lead researcher to arrange to take part.

14. What procedure is proposed for obtaining consent?

Informed consent will be ensured by participants accessing and reading the participant information sheet either electronically or via post from charities that have agreed to disseminate to their mailing lists. Participants will be asked to sign or verbally agree to consent in the research which will ensure participants are fully aware of the purpose, methods, and use of the research, and also aware of issues related to confidentiality and their right to withdraw. All participants will have received the participant information sheet and consent form at least 24 hours before the scheduled interview takes place through post or email. For face-to-face interviews the consent form will be signed immediately before the interview takes place. For participants being interviewed via Skype or telephone, participants will have the option to return the consent forms via a freepost address, or print and scan and return via email. Alternatively verbal consent can be given immediately before the interview with each item read out and verbally agreed to before the interview commences; this verbal consent will be recorded.

15. What discomfort (including psychological e.g. distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.

While the research does not anticipate to discomfort, inconvenience or danger participants, a thorough plan for this event is crucial.

In the event a participant becomes distressed, the interview will be stopped and the participant will be given all the time they need to recover and make an informed decision as to whether they would like to continue with the interview. They will be under absolutely no pressure at all to do so.

If an interview is taking place via Skype and I am not in the room with the participant, then I would look to use my clinical skills to contain the participant's distress using active listening skills and validate their feelings. I would look to ask them what support they could draw on, such as family and friends.

In the event of any risk or safeguarding concerns, professional guidelines will be followed (BPS, 2009 & HPCP, 2012). If there is a risk issue, then I would once more look to explore this risk, looking at any thoughts around harming themselves or others, plans and intent. If I was concerned regarding the safety of the participant, I would look to agree a safety plan, including the participant speaking to a friend or family member, speaking to their GP, or if necessary, presenting at a local hospital. I would agree to call/email again the following day to make sure they have accessed the support they need. Giving particular consideration to international participants, I would also provide contacts of any relevant charities that can also provide support around parenting a child with limb difference and a charity providing emotional support more broadly (Befriender Worldwide).

Following the interview, I would contact both of my research supervisors, one of which is a qualified clinical psychologist, to ensure there was no further support I could have provided. For safeguarding concerns, I would again follow professional policy and contact both of my research supervisors for further advice. For urgent risk or safeguarding concerns, I would act on this by sharing the participants address (specified on the expression of interest form) to relevant agencies for immediate support.

Participants are free to withdraw from the study at any point prior to commencing the interview without giving any reason. Once the interview has been completed, participants can withdraw their data for up to 2 weeks after the interview. After this time, the data may be analysed and incorporated into themes. Therefore it may not be possible for it to be withdrawn, though every attempt will be made to extract participants' data from the study, up until the point of publication.

16. What potential risks may exist for the researcher(s)? Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).

For face-to-face interviews, *Lancaster University's Lone Worker Guidance* will be followed. This policy addresses the health and safety requirements where persons work alone and the implementation of safe working arrangements.

Of particular relevance to the current research application:

"The worker must not leave campus without informing the Supervisor (or School) of his/her destination, nature of the work (hence hazard involved) and estimated time of return. He/she must then advise the School upon return. If the worker departs for the field directly from home, the supervisor or School must be given the relevant information by telephone and appropriate emergency plans should be in place should the lone worker fail to check in at the arranged his/her destination, nature of the work (hence hazard involved) and estimated time of return. He/she must then advise the School upon return. If the worker departs for the field directly from home, the supervisor or School must be given the relevant information by telephone and appropriate emergency plans should be in place should the lone worker fail to check in at the arranged"

As such, a 'buddy' system will also be used for any interviews at a participant's home, with a sealed envelope or password protected email given to the 'buddy' to be opened if I have not made contact with them at the agreed time. The 'buddy' will be the DClinPsy programme admin team (the 'School' in line with policy above). Inside the envelope or password protected email will be the address of the participant and my mobile number. This will only be opened in the event I had not made contact at the agreed time, thereby maintaining confidentiality whilst also taking practical steps to ensure my safety.

The '*personal safety when working off campus*' 16-point checklist to ensure safe working will also be followed to ensure the researcher's safety. Lancaster University's '*Guidance on Safety in Fieldwork*' has also been consulted, particularly the health questionnaire which satisfies the requirements to conduct field work safely while a student at the university.

Regular supervision has been agreed with my supervisors as part of the thesis contract, and therefore I would use this avenue to explore any process issues, particularly if the content of the interview was emotive. I also plan to continue to utilise my reflective journal and this has been a source of self-care throughout the course.

Any email correspondence will be from a university account and a Skype account will be set up for the purposes of this research only.

17. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.

It is hoped by participants sharing their experiences, it will help support other parents of children with limb difference by giving an understanding to and normalising some of the experiences they may encounter and the feelings associated with this. It is also hoped the results can help healthcare professionals working with parents of children with limb difference to help them understand parents' experiences, and ensure any support provided is based on parents' first hand experiences. Sharing stories is a vital part of human connection and can have an extremely powerful impact; this is one of my motivations for conducting qualitative research. Participants will also be given the option to receive a summary of the research findings should they so wish.

18. Details of any incentives/payments (including out-of-pocket expenses) made to participants:

While the study will not be offering any financial incentives, travel expenses of up to £20 per participant can be claimed. This will be at a rate of 45 pence per mile if the participant is travelling by car.

19. Briefly describe your data collection and analysis methods, and the rationale for their use. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.

The study will use a qualitative research methodology; Interpretative Phenomenological Analysis (IPA; Smith, Flowers & Larkin, 2009), or phenomenologically informed thematic analysis (Braun & Clarke, 2006). At this stage, given the sample may be international, the age of children with limb difference and the type of limb difference varied, the homogeneity of the sample is unknown, which is a requirement for the use of IPA. Therefore, having both IPA and thematic analysis as options at this stage, affords the researcher greater flexibility. If thematic analysis is used, it will draw on phenomenology to privilege the lived experience and meaning-making of parents of children with limb difference who have been provided with an artificial limb. Individual semi-structured interviews will be used to collect the data, with the questions guided by the research aims, research literature and stake-holder feedback to privilege the lived experiences of parents. Interviews will be conducted either face-to-face, via telephone or via Skype. Face-to Face interviews will be at the participant's home address.

Data used in the study will be anonymised. Interviews will be recorded using Lancaster University voice recording equipment or a Skype recorder software for interviews conducted via Skype (http://voipcallrecording.com/MP3_Skype_Recorder). The files will be stored electronically on a password protected computer and stored on the lead researcher's secure space on Lancaster University's Virtual Private Network (VPN). I will be transcribing the data with pseudonyms will be used when transcribing. Only the lead researcher, James Oliver, will have access to participant interviews, which will be stored electronically on a secure drive. The research supervisor, Dr Craig Murray, will have access to anonymised interview transcripts as part of the analysis process. Themes generated will represent the entire sample rather than specific, identifiable participants. Care will be taken not to include any quotations that may contain easily identifiable information.

Any demographic details or email addresses will be stored in a separate file from the transcripts on the university VPN, with a participant ID number assigned to identify the information.

For interviews being conducted using Skype, participants will be reminded at the start of the interview that Skype is not wholly secure, though they do have an encryption process. This point will also be on the consent form, ensuring participants are fully consenting to using Skype as a medium to take part.

A professional Skype account will be set up for the purposes of this research only and deleted at the study's conclusion.

Participants will be informed on the participant information sheet regarding the limits to confidentiality and this will also be stated on the consent forms. Should I need to break the confidentiality agreement, in the first instance I would look to contact one of my supervisors to ensure I have provided all the support I can.

20. If relevant, describe the involvement of your target participant group in the *design and conduct* of your research.

As part of the development of my research protocol, a relevant charity was consulted in the development of the interview schedule. The research idea itself was generated partly through my research supervisor's discussions with a prosthesis service in the north-west of England. Both of my research supervisors have draft read the application form and protocol and given guidance based on their respective clinical and research experience.

21. What plan is in place for the storage of data (electronic, digital, paper, etc.)? Please ensure that your plans comply with the Data Protection Act 1998.

All documents will be password protected and all data stored electronically on a secure drive (Lancaster University VPN). Physical copies of consent forms will be scanned and stored securely on the VPN. The physical consent forms will then be shredded. At the end of the study, the anonymised transcripts will be transferred electronically to the DCLinPsy Research Coordinator using a secure method supported by the University. They will be instructed with a date of when to delete the anonymised transcripts. These transcripts will be stored for 10 years before

being deleted. Files containing participant's personal/identifying details will be kept in a separate secure file from the anonymised transcripts with an ID number used to match participants identifying details to their transcripts. All personal/identifying details relating to the participants will be deleted once the thesis has been assessed.

22. Will audio or video recording take place? no audio video

If yes, what arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?

Audio recordings will be stored securely on the university VPN. Audio files will be stored until the thesis has been formally assessed. I am responsible for the deletion of audio data once the thesis has been assessed. At the end of the study, anonymised written transcripts will be transferred electronically to the DClinPsy Research Coordinator using a secure method supported by the University. These transcripts will be stored for 10 years before being deleted by the research co-ordinator.

Audio data generated from the interviews will be deleted from any portable device used as soon as possible and transferred to the secure University VPN. The reason for this is because it is not possible to encrypt the portable device. For the likely short time between the interview and transfer, the audio data on the portable device will be stored as securely as possible, and kept with the lead researcher.

23. What are the plans for dissemination of findings from the research? If you are a student, include here your thesis.

The study will be submitted as a requirement of the Doctorate in Clinical Psychology programme, specifically the doctoral thesis.

All participants in the study will be asked if they would like a copy of the full report or a summary once it has been written up and examined.

If deemed suitable, and with support of both the research and field supervisor, I will look to publish the study in a relevant peer-reviewed journal.

Finally, the research will also be presented to all year groups and research staff of the DClinPsy programme in Summer 2017.

While it is not my intention at this stage to present the findings at conference, this may change should the project be deemed of interest to any conference organisers: therefore, this remains an option.

24. What particular ethical considerations, not previously noted on this application, do you think there are in the proposed study? Are there any matters about which you wish to seek guidance from the FHMREC?

One consideration is to ensure the quotations used are not identifiable. This is a particular consideration when conducting qualitative methodology, such as IPA or thematic analysis, which may have a relatively low number of participants. Therefore, being mindful of this and using my own judgement, as well as my research supervisors, will be important.

The use of Skype/telephone interviews as a medium for conducting interviews has also been considered. Not only ethical considerations that have been covered so far on this application, such as ensuring informed consent and dealing with any possible distress or risk concerns, but also in terms of rapport and interpersonal experience. I think a key process here will be to use a reflective diary after each interview, to note down any thoughts or feelings associated with the interview and how it may differ from a face-to-face interview. Holt (2010) suggests while telephone interviews can lose some of the subtleties of communication, they can have the benefit of allowing the researcher to stay grounded to the data and avoid the imposition of contextual information. Hanna (2012) suggests Skype offers a practical solution to face-to-face interviews, particularly if video calls are utilised; this eases some (but not all) of the limitations associated with paralinguistic or interpersonal communication, whilst also offering both the participant and researcher their own safe space. It will be important to reflect on the

Research Protocol

Title: Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

Investigator Details:

Name	Contact Details
Lead researcher: James Oliver	Address: Clinical Psychology, Div. Of Health Research, Lancaster University, Lancaster, LA1 4YG Phone: [REDACTED] Email: j.oliver1@lancaster.ac.uk
Research/programme supervisor name: Dr Craig Murray	Address: Clinical Psychology, Div. Of Health Research, Lancaster University, Lancaster, LA1 4YG Phone: 01524 592730 Email: c.murray@lancaster.ac.uk
Field/Service supervisor name: Dr Clare Dixon	Address: Clinical Psychology, Div. Of Health Research, Lancaster University, Lancaster, LA1 4YG Phone: 01524 593492 Email: c.dixon3@lancaster.ac.uk

Introduction

Limb difference is an umbrella term that relates to anyone born with a congenital limb deficiency or who acquires limb loss during their life (Limbs4kids, 2015). Those with limb difference have been found to have an increased prevalence of emotional and behavioural issues and lower social competence than healthy peers, as measured through parent report on the Child Behaviour Checklist (Varni & Setoguchi, 1992). They found 23% of children were found to function in the clinically

significant range for behavioural and emotional maladjustment, with 14% of children in the social maladjustment range. The authors went on to explore the significance of these results and termed it the "new hidden morbidity" in paediatric practice. Varni & Setoguchi (1991), also explored the role of perceived physical appearance in children with congenital and acquired limb difference. They found higher perceived physical appearance was statistically predictive of lower anxiety and depression symptomology and higher self-esteem in a sample of 51 children aged between 8-13 with limb difference.

Varni and Setoguchi (1991) comment on the considerable variability in the adjustment of children with a physical disability. This variation is highlighted by a relatively recent study, that showed children with upper limb deficiency had similar levels of social competence and emotional and behavioural difficulties to a standardized norm. Interestingly, there was a significant difference between prosthesis use, with full-time users having less reported behavioural issues as assessed through questionnaire data, with this pattern stronger in girls (Hermansson, Eliasson & Engstrom, 2005).

The health-related quality of life has also been investigated in children with limb reduction deficiency (Ylimanen, Nachemson, Sommerstein, Stockselius & Hermonsson, 2010), using self-report and parent-report questionnaires. A limb reduction deficiency refers to a range of conditions that result in the arm or leg becoming shortened or incorrectly formed at birth. The study compared health related quality of life scores to a reference data set from children with other chronic health conditions. They found participants with limb reduction deficiency had a higher self-reported quality of life index score than children with other chronic health conditions. The results did show some evidence of the degree of limb loss having an impact on some aspects of health-related quality of life scores, particularly the physical limitation, emotion and social inclusion subscales.

Research related to visible difference is relevant for the current research. Children with a visible difference have been found to be at increased risk of bullying, name calling and teasing; with stigma associated with perceived disfigurement (Rumsey & Harcourt, 2007). Vannatta et al., (2009) found peer perceptions of physical appearance are correlated with peer ratings of social acceptance in a large sample of students from grades 2-10. Link and Phelan (2001) reviewed the concept of stigma, together with the relevant research literature, and suggest the impact of social stigmas can have a harmful effect on the psychosocial development of the recipient and negatively impact well-being.

Corry et al. (2009), in a review of quality of life and psychosocial adjustment to burn injury, suggest social involvement and perceived social support accounts for a significant amount of variance in psychosocial adjustment and mental health outcomes. Therefore, the role of parents in providing and facilitating social support opportunities for children may play a meaningful role in their child's adjustment. Negative social reactions to visible difference are associated with lowered self-esteem, which can lead to maintaining behaviour patterns of avoidance and safety behaviours as a way to manage this (Rumsey & Harcourt, 2007). Stock, Whale, Jenkinson, Rumsey and Fox (2013) explored young people's perceptions of visible facial differences and found while negative reactions and judgements were encountered, the authors reported they came from a lack of understanding rather than to purposefully cause harm. This is important as it perhaps shows a societal role of increasing understanding around visible difference including limb difference.

The role of parental adjustment has also been explored. Varni and Setoguchi (1993) investigated quantitatively the effects of parental adjustment on the adaptation of children with congenital or acquired limb deficiencies. Interestingly, they found parental differences, with paternal mood predicting child mood and anxiety, however maternal mood ratings did not. The relationship between parents was also found to be an important predictor, with relationship issues predicting higher levels of childhood depression, anxiety and lower self-esteem. Furthermore, family support was found to have a positive relationship to child adaptation. While the research focussed on individual parental differences, the findings support the notion of the family as a system, with that system impacting on child adaptation. The study also found the severity of the limb loss did not predict child mood, anxiety or self-esteem levels; this perhaps provides some friction to the aforementioned findings from Ylimanen et al. (2010).

Discovering that your child has a limb difference can be an emotionally poignant time for parents. Not only will there likely be an emotional impact and adjustment period for the parents and family systems, the child themselves will also have a period of adjustment. On top of this adjustment, there is a wealth of medical professionals who become involved in their child's health needs. These professionals include paediatric consultants, prosthetists, physiotherapists, occupational therapists and social workers (Limbs4life, 2013). This can add extra demands to family systems and possibly impact parental efficacy.

Morawka, Calam and Fraser (2015) comment on the increasing evidence of the importance of social environments in determining health outcomes. Parents play an integral role in managing a child's illness or disability, which has brought the focus on the role of parents in helping children to adjust to chronic health conditions or physical disabilities. Perrin et al., (1993) suggested the family environment is a key factor in contributing to a child's adjustment. According to Bronfenbrenner's theory (1986), parents are a primary source of influence to the child, with the relationship reciprocal. Therefore, the role of the parent should be acknowledged and supported where possible. Morawka et al. (2015), suggest parents need to successfully marry general parenting behaviour and specific illness management.

Research looking more generally at the impact of a child's disability on parental stress has found parental stress to be higher than parenting a child without a disability (Baker-Ericzen, Brookman-Fraze, & Stahmner, 2005). Hall, Neeley-Barnes, Graff, Krcek and Roberts (2012) suggest stress may occur through changes to the family system, with the effects potentially seen between parental relationships, the parent-child bond and relationships between siblings.

Conversely, research has also highlighted positive aspects to parenting children with a disability. Hastings, Allen, McDermott and Still (2002) have shown having a child with an intellectual disability can enhance personal growth in parents and lead to positive coping strategies. Steinton and Basser (1998) interviewed parents of a child with an intellectual disability and found a number of themes such as an increased sense of purpose, increased social network, greater joy and happiness, increased tolerance and understanding and spirituality. Of relevance to theories of adjustment and resilience, Ylven, Bjorck-Akesson and Granlund, (2006) suggest finding positive meaning to a child's disability can enhance coping and lead to a reduction in feelings of stress. In a related study, Glenn, Cunningham, Poole, Reeves & Weindling (2008) found mothers of children with Cerebral Palsy are more likely to view their emotional attachment with their child as positive when their reported stress index was low. Parents' perceptions regarding their capabilities can influence adjustment to having a child with a disability (Trute, Hieber-Murphy & Levine, 2007).

The use of a prosthesis is a key element in parenting a child with limb difference. Smith and Campbell (2009) discuss a number of key issues for parents regarding the use of an artificial limb for their child, based on their clinical experience. They suggest the nature of the limb difference is

important, with congenital limb difference more likely to result in children accepting their limb difference and less interested in an artificial limb, compared to children who experience acquired limb difference. The impact on parenting style was also commented on, with some parents being described as insisting that their child wear a prosthesis, with others choosing to give their child complete autonomy. Parental attitudes regarding the function of the prosthesis is important to consider, with some parents wanting it to look and function as the real thing, which can inadvertently communicate to the child that their limb difference is something to be ashamed of; navigating these challenges is difficult. No prior research has looked at the lived experiences of parenting a child with limb difference who have available to use a prosthesis.

The use of prostheses' in adults has received some attention in the past. Of particular relevance is the work of Murray (2010), who discussed both the personal meanings and social meaning of prosthesis use. Murray suggests for some the personal meaning attributed to using a prosthesis goes beyond the functional capabilities of the device, and is entangled with more significant considerations that it brings, such as perceiving to be living a life like others, treated like others and enabling independence and social participation. Of particular importance to the current study, Murray discussed the social meanings afforded to prosthesis use, with social response playing an important role in prosthesis use. The current study will look to add to this interplay between the child's social system, and the perceived views of the parental system to see if it may influence prosthesis use.

There have been a number of theoretical models developed to provide a framework for understanding the research evidence that children with a chronic illness or disability are at increased risk of developing psychological difficulties. Some of the models have focussed at an individual level of adaptation, such as Wallander and Varni's Disability Stress Coping Model (1992) or the Transaction Coping and Stress model (Thompson & Gustafson, 1996). While both of these models incorporate family processes, other models have focussed more attention to family adaptation such as The Resiliency Model of Family Stress, Adjustment, and Adaptation (McCubbin & McCubbin, 1993), the Family Management Style Framework (Knafl & Deatrick, 2003) and the Family Systems Illness Model (Rolland, 1994).

Both attachment theory (Bowlby, 1969), attachment types (Ainsworth, Blaher, Waters & Wall, 1978) and Erikson's (1968) psychosocial stage model of identity will also be considered in the proposed

research. Understanding parental experiences may give insight into demands or resources that have relevance to attachment theory and the development of a secure and safe base for their child. Erikson's psychosocial theory of development may also be an importance consideration, particularly given the age range of the children included in the study; 5-16 years of age. Systemic family theory is another important consideration, with the family context influencing the way the family system views the world (Dallos & Draper, 2005). Models of adjustment related to parent experiences (e.g. Wallander & Varni, 1992) and models of resilience taken from positive psychology (Synder & Lopez, 2009) will also be considered to see if these psychological theories can inform the research findings. Benefit finding and growth may also be a consideration which may help move away from a traditional medical model of disability, focussing on the limitations, toward a more holistic psychological approach drawing out parental resources and the positives of parenting a child with limb difference.

Given the issues discussed, an exploration of the experiences of being a parent of a child with limb difference, who use, or have available to use, an artificial limb, would be of benefit to parents adjusting to their child's limb difference and also to healthcare professionals working within paediatric settings. It could help provide insight into the lived experiences of parents and give insight into some of the challenges that need to be navigated by parents of children with limb difference. It would be of particular relevance to clinical psychology and theories of adjustment, attachment and systemic family theory in understanding any barriers or facilitators to interventions and enable consideration of a psychological perspective in a medical Multi-Disciplinary Team (MDT). The research may also identify additional support or resources that would help parents to support their child, and give parents a voice to talk about their experiences in a rich narrative.

The research question is 'What are the experiences of parents of children with limb difference who have been provided with an artificial limb, and how do they make sense of these experiences?' The research aims are (i) to develop an understanding of the experiences of being a parent of a child with limb difference who has been provided with an artificial limb, (ii) to develop an understanding of how these experiences are understood by parents and managed, (iii) to consider the role of psychological theory in understanding these experiences, and (iv) to consider how clinical psychologists working within paediatric health settings can use their skills to support parents and inform clinical service structure.

Method

Design

The study will use a qualitative research methodology; Interpretative Phenomenological Analysis (IPA; Smith, Flowers & Larkin, 2009), or phenomenologically informed thematic analysis (Braun & Clarke, 2006). At this stage, given the sample may be international, the age of children with limb difference and the type of limb difference varied, the homogeneity of the sample is unknown, which is a requirement for the use of IPA. Therefore, having both IPA and thematic analysis as options at this stage, affords the researcher greater flexibility. If thematic analysis is used, it will draw on phenomenology to privilege the lived experience and meaning-making of parents of children with limb difference who have been provided with an artificial limb. Individual semi-structured interviews will be used to collect the data, with the questions guided by the research aims, research literature and stake-holder feedback to privilege the lived experiences of parents.

Participants

Participants will be parents of children with limb loss. The specific inclusion and exclusion criterion are as follows:

Inclusion:

- Parents to have a child with a limb difference and have available to use an artificial limb.
- Age range of child to be between 5-16 years old (primary and secondary school).
- Child's limb difference can be congenital or acquired through any event e.g. trauma or resultant from a health condition such as meningitis.
- Child to have had the availability of a prosthesis for a minimum of 6 months.
- Parents to speak English.

Note on Inclusion:

- Rationale for this age range is that I want to capture parental experiences across key developmental milestones and important life events (e.g. having begun attending school, transition to high school, developing identity through puberty). The idea is to keep the age range broad because the research is novel and exploratory.

- In the event the study is oversubscribed, I will use a quota to choose particular participants which would provide the study with variety, such as an equal split of genders or particular age ranges of children e.g. x amount of 7-10 year olds.
- If the study is not oversubscribed, then I will include all participants who meet the inclusion criteria until the recruitment target is achieved.

Exclusion:

- Parents of children with a co-morbid health condition which may limit or preclude prosthesis use or any other physical and/or intellectual disability.

The rationale for this is that the presence of any of these factors may mean parents are facing different challenges and experiences to those posed specifically by having a child with limb difference, who have been provided with an artificial limb.

Sample

A sample size of between 4-12 participants are anticipated to be recruited to the proposed study. The sample will be international. Sample size is often a contentious issue in qualitative research. Sandelowski (1995) suggests the sample needs to be large enough to gain a rich understanding of the data, while small enough to manage the transcript material.

The study is aiming to explore parents' experiences of having a child with limb difference who have been provided artificial limb. A semi-structured interview based on the identified gaps in the research literature and project aims will be conducted (see appendix C for an example interview schedule). The interview schedule will be semi-structured allowing flexibility from the interviewer to respond appropriately to the participant. The interviews are anticipated to last around one hour.

Recruitment

Participants will be recruited via online charities. Recruitment will take the form of a three-point plan where possible:

1. *Via relevant and amiable charities advertising and disseminating the participant information sheet via social media, home webpages and mailing lists*

Relevant charities will be contacted via phone and/or email (see appendix D for draft email correspondence). Charities will be emailed the participant information sheet (appendix A) and the social media advertisement with an electronic link to the participant information sheet (appendix E), and asked to share this via their home webpage, Twitter and Facebook accounts. Furthermore, charities will also be asked if they are able to send the participant information sheet to their mailing lists. Finally, charities will also be asked to advertise the study in any publications they have and then sent an advertisement if they are happy to do so (appendix F).

2. Via Twitter

The lead researcher proposes to set up a research twitter account that can have a link to the participant information sheet (on a research advertising space for DClinPsy students). The Twitter account would be solely used to advertise the proposed study and to share the participant information sheet. Active online charities will be contacted and asked to consider 're-tweeting' my post (see appendix E), so other followers of that site can voluntarily gain access to the study information and email the lead researcher to take part.

3. Via Facebook

Active charities will be asked to share the participant information sheet on their Facebook page. No specific Facebook account will be set up by the lead researcher, rather the participant information sheet will be sent to the charities and they will be asked to share the link to the electronic version on their Facebook page. Again, participants can access the study details and get in touch with the lead researcher to arrange to take part.

Proposed analysis

The interview data will be analysed using IPA or phenomenologically informed thematic analysis. Both methods are a form of thematic analysis and in essence involve close analysis of the interview transcripts, coding of the material, before the generation of broader overarching themes that capture the essence of the data. IPA will use the guidelines from Smith, Flowers and Larkin (2009), with Braun and Clarke's (2006) guidelines used if using phenomenologically informed thematic analysis. A clear audit trail will be generated which will explicitly highlight the coding process, so it is clear where the

themes emerged from. The research supervisor will also be involved in the analysis which will act as a form of rigour and validity, to ensure a form of quality assurance to the analysis.

Procedure

Participants are to be recruited through charities supporting both children and adults with limb difference. A number of leading international charities have been tentatively approached by the lead researcher, James Oliver, and the research supervisor Dr Craig Murray. These charities have provisionally agreed to advertise the study once it has gained ethical approval. Given the lack of research exploring parental experiences, a number of charities are understandably enthusiastic regarding the proposed research. These charities have all agreed to advertise the study via their social media sites and some have agreed to advertise the study in publications. The charities will advertise the participant information sheet which will contain an email contact and a UK mobile number to contact should they wish to take part. Once contacted, the lead researcher will request participants to fill in an expression of interest form (see appendix H), which will ask for details such as the age and gender of the participant's child, confirmation that they have had available to use a prosthesis for a minimum of 6 months, and their preference for the interview medium e.g. face-to-face, telephone or Skype. The participant's country of origin and address will also be requested, which will ensure adequate procedures can be followed in the event of any risk or safeguarding concerns, in particular the sharing of participant's address with relevant authorities should an immediate risk present. Following completion of the expression of interest form, the lead researcher will arrange the interview in-line with the expressed preferences.

All participants will receive the consent form at least 24hours before the scheduled interview takes place through post or email. For face-to-face interviews the consent form will be signed immediately before the interview takes place. For participants being interviewed via Skype or telephone, participants will have the option to return the consent forms via a freepost address, or print and scan and return via email. Alternatively verbal consent can be given immediately before the interview with each item read out and verbally agreed to before the interview commences; this verbal consent will be recorded.

For face-to-face interviews, *Lancaster University's Lone Worker Guidance* will be followed. As such, a 'buddy' system will also be used for any interviews at a participant's home, with a sealed envelope or password protected email given to the 'buddy' to be opened if I have not made contact with them at the agreed time. Inside the envelope will be the address of the participant, this will only be opened in the event I had not made contact at the agreed time, thereby maintaining confidentiality whilst also taking practical steps to ensure my safety. All interviews will be audio recorded and later transcribed by the lead researcher. All data will remain confidential. Data used in the study will be anonymised. Interviews will be recorded using Lancaster University voice recording equipment. If using Skype, interviews will also be recorded by a software allowing Skype calls to be audio recorded (www.voipcallrecording.com/MP3_Skype_Recorder). The recordings are an MP3 file that will be stored securely on Lancaster University Virtual Private Network (VPN) as soon as the interview has taken place. Files will be stored electronically on the VPN which is a password protected, encrypted drive. Digital audio files will be stored onto the VPN and deleted from the recorder as soon as they have been transcribed. The digital audio files will be deleted from the secure drive once the thesis has been formally assessed. Anonymised electronic transcriptions will also be stored on the VPN. Anonymised electronic transcripts are stored at Lancaster University for up to 10 years after the completion of the study.

Materials

A semi-structured interview will be used; see appendix C for example. A participant information sheet (appendix A) will be advertised via agreeable limb difference related charities, and also advertised via the lead researcher's Twitter account. This account will be created once ethical approval has been granted and will only be used for the purpose of advertising the proposed study. Upon completion of the study the Twitter account will be deleted.

At the end of the study, participants will be debriefed (appendix G), this debrief will give relevant contact numbers for further support should it be required.

Stake-Holder and Service User Liaison

The interview schedule has been shared with a charity supporting parents of children with limb difference and was also shared with a parent of a child with limb difference who is part of the charity's committee. They agreed to view and provide feedback to the interview schedule

Practical issues (e.g., costs/logistics)

There will be costs associated with international calls via Skype. These costs have been given approval by the DClinPsy Programme Director, Professor Bill Sellwood. Where possible face-to-face interviews will take place, however this is unlikely to be always possible given the international sample. Where relevant, reasonable travel expenses will be offered to participants up to maximum of £20.

Ethical Considerations

Informed consent

Informed consent will be ensured by participants accessing and reading the participant information sheet either electronically or via post from charities that have agreed to disseminate to their mailing lists. Participants will be asked to sign or verbally agree to consent in the research which will ensure participants are fully aware of the purpose, methods, and use of the research, and aware of issues related to confidentiality and their right to withdraw. All participants will have received the participant information sheet and consent form at least 24hours before the scheduled interview takes place through post or email. For face-to-face interviews the consent form will be signed immediately before the interview takes place. For participants being interviewed via Skype or telephone, participants will have the option to return the consent forms via a freepost address, or print and scan and return via email. Alternatively verbal consent can be given immediately before the interview with each item read out and verbally agreed to before the interview commences; this verbal consent will be recorded.

Confidentiality

All data will remain confidential. However, there are some limits to confidentiality; specifically, if something said in the interview indicates either the participant, or another, may be at risk of harm. In which case, the lead researcher has a duty of care to pass on that information to relevant agencies. Where possible I would inform the participant of my intention to share the

information. Data used in the study will be anonymised. Interviews will be recorded using Lancaster University voice recording equipment or a Skype recorder for interviews conducted via Skype. Audio recordings will be stored securely on the university VPN. Audio files will be stored until the thesis has been formally assessed. I am responsible for the deletion of audio data once the thesis has been assessed. Pseudonyms will be used when transcribing the data, with the transcriptions completed by the lead researcher. At the end of the study, anonymised written transcripts will be transferred electronically to the DClinPsy Research Coordinator using a secure method supported by the University. These transcripts will be stored for 10 years before being deleted by the research coordinator.

Participants are free to withdraw from the study at any point prior to commencing the interview without giving any reason. Once the interview has been completed, participants can withdraw their data for up to 2 weeks after the interview. After this time, the data may be analysed and incorporated into themes. Therefore it may not be possible for it to be withdrawn, though every attempt will be made to extract participants' data from the study, up until the point of publication. Only the lead researcher, James Oliver, will have access to participant interviews, which will be stored electronically on a secure drive (VPN). The research supervisor, Dr Craig Murray, will have access to anonymised interview transcripts as part of the analysis process. Themes generated will represent the entire sample rather than specific, identifiable participants. Care will be taken not to include any quotations that may contain easily identifiable information. For interviews being conducted using Skype, participants will be reminded that Skype is not completely secure though they do have an encryption process. This point will also be on the consent form, ensuring participants are fully consenting to using Skype as an interview medium.

Potential Distress

In the event a participant becomes distressed, the interview will be stopped and the participant will be given all the time they need to recover and make an informed decision as to whether they would like to continue with the interview. They will be under absolutely no pressure at all to do so.

If an interview is taking place via Skype and I am not in the room with the participant, then I would look to use my clinical skills to contain the participant's distress by using active listening skills and looking to validate their feelings. I would look to ask them what support they could draw on such as family, friends and sources of online support such as Befrienders Worldwide (www.befrienders.org)

In the event of any risk or safeguarding concerns, professional guidelines will be followed (BPS, 2009 & HPCP, 2012). If there is a risk issue, then I would once more look to explore this risk, looking at any thoughts around harming themselves or others, plans and intent. If I was concerned regarding the safety of the participant, I would look to agree a safety plan, including the participant speaking to a friend or family member, speaking to their GP, or if necessary, presenting at a local hospital. I would agree to call/email again the following day to make sure they have accessed the support they need. Giving particular consideration to international participants, I would also provide contacts of any relevant charities that can also provide support around parenting a child with limb difference and a charity providing emotional support more broadly (Befriender Worldwide). Following the interview, I would contact both of my research supervisors, one of which is a qualified clinical psychologist, to ensure there was no further support I could have provided. For safeguarding concerns, I would again follow professional policy and contact both of my research supervisors for further advice. For urgent risk or safeguarding concerns, I would act on this by sharing the participants address (specified on the expression of interest form) to relevant agencies for immediate support.

Withdrawal

Participants are free to withdraw from the study at any point prior to commencing the interview without giving any reason. Once the interview has been completed, participants can withdraw their data for up to 2 weeks after the interview. After this time the data will be analysed and incorporated into themes. Therefore, it may not be possible for it to be withdrawn, though every attempt will be made to extract participants' data from the study, up until to the point of publication.

Study Oversubscription

In the event the study is oversubscribed, I will approach the issue with sensitivity. I would explain to potential participants that the study is oversubscribed and a quota was used to ensure a

variety within the sample. I would offer them a summary of the results or a full copy of the paper once the study is complete.

Timescale

The proposed research is part of the DCLinPsy thesis and is required to be submitted to Lancaster University by the middle of May 2017. It is anticipated participants will be interviewed once ethical approval is granted. It is anticipated this stage will be from August 2016 to December 2016.

If it is decided the research will be submitted for publication this is likely to happen by October 2017.

All participants will receive a copy or summary of the research paper by 31st August 2017 if requested.

References

- Ainsworth, M. D. S., Blehar, M. C., Waters, E., & Wall, S. (1978). *Patterns of attachment: A psychological study of the strange situation*. Hillsdale, NJ: Erlbaum
- Akinsola, E. F., & Udoka, P. A. (2013). Parental influence on social anxiety in children and adolescents: Its assessment and management using psychodrama. *Psychology, 4*, 246-253. doi:10.4236/psych.2013.43A037
- Baker-Ericzén, M. J., Brookman-Frazee, L., & Stahmner, A. (2005). Stress levels and adaptability in parents of toddlers with and without autism spectrum disorders. *Research & Practice for Persons with Severe Disabilities, 30*(4), 194-204. doi:10.2511/rpsd.30.4.194
- Bowlby J. (1969). *Attachment. Attachment and loss: Vol. 1. Loss*. New York: Basic Books.
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology, 3*(2), 77-101. doi:10.1191/1478088706qp063oa
- British Psychological Society (2009). Code of Ethics and Conduct Guidance published by the Ethics Committee of the British Psychological Society. Retrieved from http://www.bps.org.uk/system/files/Public%20files/bps_code_of_ethics_2009.pdf
- Bronfenbrenner, U. (1979). *The ecology of human development: Experiments by nature and design*. Cambridge, MA: Harvard University Press.
- Corry, N., Pruzinsky, T., & Rumsey, N. (2009). Quality of life and psychosocial adjustment to burn injury: Social functioning, body image, and health policy perspectives. *International Review of Psychiatry, 21*(6), 539-548. doi:10.3109/09540260903343901
- Dallos, R., & Draper, R. (2005). *An Introduction to Family Therapy: Systemic Theory and Practice*. Maidenhead: Open University Press.
- Erickson, E. (1968). *Identity, Youth and Crisis*. New York: Norton.
- Glenn, S., Cunningham, C., Poole, H., Reeves, D., & Weindling, M. (2008). Maternal parenting stress and its correlates in families with a young child with cerebral palsy. *Child care, health and development, 35*, 71-78. doi: 10.1111/j.1365-2214.2008. 00891.x
- Hall, H. R., Neely-Barnes, S. L., Graff, J. C., Krcek, T. E., Roberts, R. J., & Hankins, J. S. (2012). Parental stress in families of children with a genetic disorder/disability and the resiliency

- model of family stress, adjustment, and adaptation. *Issues in Comprehensive Pediatric Nursing*, 35(1), 24-44. doi:10.3109/01460862.2012.646479
- Hanna, P. (2012). Using internet technologies (such as Skype) as a research medium: A research note. *Qualitative Research*, 12(2), 239-242. doi:10.1177/1468794111426607
- Hastings, R. P., Allen, R., McDermott, K., & Still, D. (2002). Factors related to positive perceptions in parents of children with intellectual disabilities. *Journal of Applied Research in Intellectual Disabilities*, 15(3), 269-275.
- Health and Care Professionals Council (2012). Standards of conduct, performance and ethics. Retrieved from <http://www.hcpc-uk.org.uk>
- Hermansson, L., Eliasson, A., & Engström, I. (2005). Psychosocial adjustment in Swedish children with upper-limb reduction deficiency and a myoelectric prosthetic hand. *Acta Paediatrica*, 94(4), 479-488. doi:10.1080/0803525041002251
- Holt A (2010) Using telephones for narrative interviewing: a research note. *Qualitative Research*, 10, 113-121. doi: 10.1177/146879410934868
- Knafl, K. A., & Deatrick, J. A. (2003). Further refinement of the family management style framework. *Journal of Family Nursing*, 9(3), 232-256. doi:10.1177/1074840703255435
- Limbs4life (2013). *Information for parents and carers of a child with limb difference*. Retrieved from: www.limbs4life.org.au/.../Limbs4Life-CCY-Parents-Handbook-WEB.pdf
- Limbs4kids (2015). *Fact Sheet 1: About Limb Difference and Limb Difference Professionals*. Retrieved from: <http://limbs4kids.org.au/wp-content/uploads/2015/05/Fact-Sheet-1-About-Limb-Difference.pdf>
- Link, B. G., & Phelan, J. C. (2001). Conceptualizing stigma. *Annual Review of Sociology*, 27, 363-385. doi: 10.1146/annurev.soc.27.1.363
- Morawska, A., Calam, R., & Fraser, J. (2015). Parenting interventions for childhood chronic illness: A review and recommendations for intervention design and delivery. *Journal of Child Health Care*, 19(1), 5-17. doi:10.1177/1367493513496664
- McCubbin, M. A., & McCubbin, H. I. (1993). Families coping with illness: The Resiliency Model of Family Stress, Adjustment, and Adaptation. In C.B. Danielson, B. Hamel- Bissell, & P.

- Winstead-Fry (Eds.), *Families, health, and illness: Perspectives on coping and intervention* (pp. 21-63). St. Louis: Yearbook
- Murray, C. (2010). Understanding Adjustment and Coping to Limb Loss and Absence through Phenomenologies of Prosthesis Use. In C. Murray (Eds.), *Amputation, Prosthesis Use, and Phantom Limb Pain. An Interdisciplinary Perspective* (pp. 81-100). London: Springer
- Perrin, E. C., Ayoub, C. C., & Willett, J. B. (1993). In the eyes of the beholder: Family and maternal influences on perceptions of adjustment of children with a chronic illness. *Journal of Developmental and Behavioral Pediatrics, 14*(2), 94-105. Retrieved from PsychINFO.
- Rolland, J.S. (1994a) *Families, Illness, and Disability: An Integrative Treatment Model*. New York: Basic Books.
- Rumsey, N. & Harcourt, D. (2007). Visible difference amongst children and adolescents: Issues and interventions. *Developmental Neurorehabilitation, 10*, 113-123.
doi:10.1080/13638490701217396
- Sandelowski, M. (1995). Sample size in qualitative research. *Research in Nursing & Health, 18*, 179-183. doi:10.1002/nur.4770180211
- Smith, D. G. & Campbell, K. M. (2009). Prosthesis for children and teens with limb difference: Issues and expectations. *InMotion, 19*(2) 12-18. Retrieved from http://www.amputee-coalition.org/inmotion/mar_apr_09/prostheses_for_children.html
- Smith, J. A., Flowers, P., & Larkin, M. (2009). *Interpretative phenomenological analysis: Theory, method and research*. London: Sage.
- Stainton, T., & Besser, H. (1998). The positive impact of children with an intellectual disability on the family. *Journal of Intellectual and Developmental Disability, 23*(1), 57-70.
doi:10.1080/13668259800033581
- Stock, N., Whale, K., Jenkinson, E., Rumsey, N. & Fox, F. E. (2013) Young people's perceptions of visible difference. *Diversity and Equality in Health and Care, 10* (1). 41-51. Retrieved from: <http://eprints.uwe.ac.uk/21144>
- Stein, R. E. K., & Jessop, D. J. (1989). What diagnosis does not tell: The case for a non-categorical approach to chronic illness in childhood. *Social Science and Medicine, 29*, 769-778.
doi:10.1016/0277-9536(89)90157-3

- Synder, C. R., & Lopez, S. J. (2009). *Handbook of Positive Psychology (2nd edition)*. New York: Oxford University Press
- Thompson, R. J., & Gustafson, K. E. (1996). *Adaptation to chronic childhood illness*. Washington, DC: American Psychological Association.
- Trute, B., Hiebert-Murphy, D., & Levine, K. (2007). Parental appraisal of the family impact of childhood developmental disability: Times of sadness and times of joy. *Journal of Intellectual and Developmental Disability, 32(1)*, 1-9. doi:10.1080/13668250601146753
- Vanatta, K. Gartstein, M. A., Zeller, M. & Noll, R. B. (2009). Peer acceptance and social behaviour during childhood and adolescence: How important are appearance, athleticism and academic competence? *International Journal of Behavioural Development, 33*, 303-311. doi:10.1177/0165025408101275
- Varni, J. & Setoguchi, Y. (1991). Correlates of perceived physical appearance in children with congenital/acquired limb deficiencies. *Journal of Developmental and Behavioral Pediatrics, 12*, 171-176. doi:10.1097/00004703-199106000-00005
- Varni, J. W., & Setoguchi, Y. (1992). Screening for behavioral and emotional problems in children and adolescents with congenital or acquired limb deficiencies. *American Journal of Diseases of Children (1960), 146(1)*, 103-107. Retrieved from MEDLINE
- Varni, J. W., & Setoguchi, Y. (1993). Effects of parental adjustment on the adaptation of children with congenital or acquired limb deficiencies. *Journal of Developmental and Behavioral Pediatrics, 14(1)*, 13-20. doi:10.1097/00004703-199302000-00003
- Wallander, J. L., & Varni, J. W. (1992). Adjustment in children with chronic physical disorders; Programmatic research on a disability-stress-coping model. In A. M. LaGreca, L. Siegal, J. L. Wallander, & C. E. Walker (Eds.), *Stress and coping with pediatric conditions* (pp. 279-98). New York: Guilford Press.
- Wallander, J.L., & Varni, J.W. (1998). Effects of paediatric chronic physical disorders on child and family adjustment. *Journal of Child Psychology and Psychiatry, 39(1)*, 29-46. doi: 10.1111/14697610.00302

- Ylimainen, K., Nachemson, A., Sommerstein, K., Stockslius, A., & Hermansson, L. N. (2010). Health-related quality of life in Swedish children and adolescents with limb reduction deficiency. *Acta Paediatrica*, *99*(10), 1550-1555. doi:10.1111/j.1651-2227.2010.01855.x
- Ylven, R., Bjorck-Akesson, E., & Granlund, M. (2006). Literature review of positive functioning in families with children with a disability. *Journal of Policy and Practice in Intellectual Disabilities*, *3*, 253-270. doi:10.1111/j.1741-1130.2006.00089.x

Appendix A: Participant Information Sheet

Title: *Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.*

Hello my name is James Oliver and I am a trainee clinical psychologist. I am conducting this research as part of my studies for a Doctorate in Clinical Psychology at Lancaster University, Lancaster, United Kingdom. Thank you for taking the time to read this information sheet and **if at the end of reading the information you would like to take part, or would like further information, then please do not hesitate to contact me at j.oliver1@lancaster.ac.uk**

What is the study about?

The study is looking to interview parents of children with limb difference aged between 5-16 years old who use, or have available to use, an artificial limb. There is little research exploring parents lived experiences of parenting a child with limb difference, and little regarding the prosthetic rehabilitation process. Therefore, this study is looking to address this gap, and give a platform for parents to speak about their experiences. To take part your child must have had the availability of a prosthesis for a minimum of 6 months.

What will I be asked to do if I take part?

You will be asked to take part in an interview lasting approximately 45 minutes to an hour with the lead researcher, James Oliver. This interview will be audio recorded and you will be asked about your experiences. These experiences will be unique to you, but as a general guide may involve questions exploring your parenting experiences, the prosthetic rehabilitation process, any challenges you have encountered, and how you have managed and made sense of these experiences.

This interview will be arranged with you and can be completed via Skype, telephone or possibly in person if based in the North-West of England, UK. Please note that Skype interviews are not wholly secure due to the nature of the platform. However, Skype have an encryption process in place and further information around Skype's security can be found at:
<https://www.skype.com/en/security/#encryption>

Do I have to take part?

No you are under no obligation to take part.

Will my data be Identifiable?

The information you provide is confidential and all data collected will be anonymised. The typed version of your interview will be made anonymous by removing any identifying information including your name. A pseudonym (false name) will then be used for any direct quotations used in the write-up of the study.

The data collected for this study will be stored securely. Only the lead researcher, James Oliver, will have access to participant interviews, which will be stored electronically on a secure drive. The research supervisor, Dr Craig Murray, will have access to anonymised interview transcripts. All files relating to the study will be password protected and encrypted and kept on Lancaster University's secure server to ensure confidentiality. At the end of the study, electronic copies of anonymised transcripts will be kept securely for 10 years at Lancaster University in line with university policy. At the end of this period, they will be deleted. Audio recordings will be deleted once the project has been submitted for publication/examined.

There are some limits to confidentiality; specifically, if something said in the interview indicates that you or someone else may be at risk of harm. In which case, the lead researcher has a duty of care to pass on that information to relevant agencies to ensure your or the person's safety. If possible, I will tell you if I have to do this.

What will happen to the results?

The results will be written up into a research paper and will be assessed as part of the lead researcher's Doctorate in Clinical Psychology qualification. It may be decided to submit the report to

a peer-reviewed journal, which if published, can be accessed via the internet. You will be offered a copy of the final research paper or summary of the findings once they have been written, to see how your input contributed to the findings.

Can I withdraw from the study?

Your participation in the study is voluntary and you are free to withdraw at any point prior to commencing the interview without giving any reason. Once the interview has been completed, you can withdraw your data for up to 2 weeks after the interview. After this time, the data may be analysed and incorporated into themes. Therefore it may not be possible for it to be withdrawn after this time.

Are there any risks?

There are no risks anticipated with participating in this study. However, if you experience any distress following participation, you are encouraged to inform the researcher. The debrief sheet also contains resources which can provide further support.

Are there any benefits to taking part?

There are no direct benefits to you for taking part. However, it is hoped by sharing your experiences it will help support other parents of children with limb difference, by giving an understanding of some of the experiences they may encounter and the feelings associated with this. It is also hoped the results can help healthcare professionals working with the parents of children with limb difference, to help them understand parent's experiences, and ensure any support provided is based on parents first hand experiences. If required, reasonable travel expenses will be paid up to a maximum of £20.

Who has reviewed the project?

This study has been reviewed by the Faculty of Health and Medicine Research Ethics Committee, and approved by the University Research Ethics Committee at Lancaster University.

Where can I obtain further information about the study if I need it?

James Oliver

Doctorate in Clinical Psychology

Furness College

Lancaster University

Lancaster

LA1 4YG

Email: j.oliver1@lancaster.ac.uk UK Research Mobile number: + [REDACTED]

Complaints

If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:

Professor Bruce Hollingsworth
Head of Department of the Div. of Health Research
Lancaster University
Lancaster
LA1 4YG
Tel: 01524 594154
Email: b.hollingsworth@lancaster.ac.uk

Professor Bill Selwood
Programme Director DClinPsych
Lancaster University
Lancaster
LA1 4YG
Tel: 01524 593998
Email: b.sellwood@lancaster.ac.uk

Professor Roger Pickup
Associate Dean for Research
Faculty of Health and Medicine (Division of Biomedical and Life Sciences),
Lancaster University
Lancaster
LA1 4YD
Email: r.pickup@lancaster.ac.uk Tel: 01524 593746

Thank you for taking the time to read this information sheet.

Appendix C: Example Semi-Structured Interview

Demographic Information

Before commencing the interview could you briefly tell me how old your child is and the nature of their limb difference?

Parenting

What have been the key challenges or adaptations to parenting a child with limb difference?

How have you managed these challenges?

Have there been any benefits?

Has there been any impact to your family system? If so, what are some of the difficulties or positives encountered? Who, if anyone, in the family has been impacted?

How have you managed your own needs when parenting a child with limb difference?

Limb difference and prosthesis use

What was your understanding of limb difference and prosthesis use before you had your child?

What does it mean to you now?

How did you decide for your child to be provided with an artificial limb?

Did you seek support in this decision making process (family, friends, healthcare professionals)? If so what is your experience of receiving this?

How would you describe your relationship with the healthcare services supporting your child? How have you experienced this support?

Have there been any disagreements with healthcare professionals involved in your child's artificial limb use? If so, how have these been managed? If not, why do you think that is?

Prosthesis use and parenting

How has your child adapted to using an artificial limb?

Has this brought any challenges in your role as a parent?

How do you think your child feels about their artificial limb? What experiences have you had that has made you think this?

Have there been any benefits to your child in using an artificial limb?

Do you think your child has been impacted in anyway by having a limb difference? (school, friendships). Has using an artificial limb changed this in anyway?

Final thoughts

If you had to give advice for new parents of a child with limb difference what would it be?

If you could give advice or make any changes to the healthcare services involved during your journey of parenting a child with limb difference what would it be?

Specifically, to artificial limb use, what advice would you give to prosthesis services, to consider parents' experiences?

Thank you for taking part, do you have any final thoughts or reflections about your experiences of parenting a child with limb difference?

Appendix D: Covering Email to Charities/Advocacy Groups

James Oliver
E-mail - j.oliver1@lancaster.ac.uk
Telephone [REDACTED]
DClinPsy Lancaster University
Clinical Psychology
Division of Health Research
Lancaster
LA1 4YG

Dear [charity/advocacy group]

Firstly, I would like to introduce myself. My name is James Oliver and I am currently completing a Doctorate in Clinical Psychology at Lancaster University, UK. As part of the programme, we conduct research into a topic area of our choice. In collaboration with Dr Craig Murray (Research Supervisor), we are hoping to look at a valuable and under-research topic area:

Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

As part of this research, I will be looking to interview parents of children with limb difference who have been through the prosthetic rehabilitation process with their child. To date, there is very little research relating to how parents experience their parenting role and the prosthetic rehabilitation process. It is hoped the research can give a voice to parents of children with limb difference and help other parents who may be starting a similar journey. It is also hoped the research will help inform healthcare services to best support parents, by understanding some of the challenges and experiences of parenting a child with limb difference.

The research will involve conducting either a Skype/telephone/or face-to-face interview with myself, James Oliver, the lead researcher. These interviews will be audio recorded and later transcribed with all identifying information anonymised to maintain participant's confidentiality. I have attached a participant information sheet and a social media advertisement for Twitter or Facebook. The participant information sheet can also be accessed electronically:
(add URL link to participant information sheet).

In the first instance, I would be delighted if you could advertise the study via your webpage, Twitter or Facebook accounts, where potential participants will be able to click on the electronic link to access the study information and to get in contact should they wish to take part.

Alternatively, if you think it may be possible for you to send the participant information sheet to anyone on your mailing list who you think may be interested, that would be very much appreciated.

I am hoping to interview approximately 12 participants. I really hope to achieve this and in doing so make the research a valuable contribution to an under-researched area. The aim would be to publish this research to add to the evidence base regarding how health related services can best support parents.

I look forward to your reply and if you have any further questions or queries please do not hesitate to contact me at j.oliver1@lancaster.ac.uk or alternatively I can ring at a convenient time to suit you.

Kind regards

James Oliver
Trainee Clinical Psychologist
Lancaster University

Appendix E: Social Media Advertisement

Twitter (max 140 characters)

Option A

Are you the parent of a child with limb difference who has been provided with an artificial limb? Research opportunity...
(electronic link to Participant Information Sheet)

Option B

Are you the parent of a child with limb difference? Opportunity to be involved in research
(electronic link to Participant Information Sheet)

Option C

Are you the parent of a child with limb difference who has been provided with an artificial limb? Have your say...
(electronic link to Participant Information Sheet)

Facebook- Option A

Are you the parent of a child with limb difference who has been provided with an artificial limb? Opportunity to be involved in research and have your say ...
(electronic link to Participant Information Sheet)

Facebook B - Option B

Are you the parent of a child with limb difference aged 5-16 who has been provided with an artificial limb? Opportunity to be involved in research and have your say ...
(electronic link to Participant Information Sheet)

Appendix F: Expression of Interest to be Advertised in Relevant Charity Publications

Are you the parent of a child with limb difference who has been provided with an artificial limb? Opportunity to be involved in research and have your say...

I'm researching the experiences of being a parent of a child with limb difference who has been provided with an artificial limb. The study will involve an interview with myself, James Oliver (Trainee Clinical Psychologist). It will involve talking about your parenting experiences and your experiences of the prosthetic rehabilitation process for your child. I am inviting participants from around the world to take part. As a result, the interview can be completed via Skype, telephone or in person depending on where you reside. Data used in the study will be anonymised, with a pseudonym (false name) used for any direct quotations used in the write-up of the study.

If you would like to take part or obtain further information, then please do not hesitate to get in touch at j.oliver1@lancaster.ac.uk or tweet at (twitter address).

I look forward to hearing from you,

James Oliver
Trainee Clinical Psychologist
Lancaster University



Appendix G: Debrief

James Oliver
E-mail - j.oliver1@lancaster.ac.uk
Telephone [REDACTED]
DClinPsy Lancaster University
Clinical Psychology
Division of Health Research
Lancaster
LA1 4YG
Research Supervisor- Dr Craig Murray
E-mail - C.Murray@lancaster.ac.uk

Thank you for taking part in this study. The study was looking to explore the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

If you have any questions or concerns relating to the study please contact the lead researcher, James Oliver, in the first instance at j.oliver1@lancaster.ac.uk. If you have any concern or complaints regarding the study please contact Professor Bruce Hollingsworth at b.hollingsworth@lancaster.ac.uk, or Professor Bill Selwood at b.sellwood@lancaster.ac.uk, or Professor Roger Pickup, Associate Dean for Research at r.pickup@lancaster.ac.uk.

You may wish for further resources relating to the research, and as you may be aware, there are a number of international charities that can be contacted for further support and information. I have listed some of the charities I am aware of and have been in touch with regarding this research process.

- <http://www.limbless-association.org/>
- <http://www.limbs4life.org.au/>
- <http://limbs4kids.org.au/>
- <http://www.dysnet.org/>
- <http://www.amputee-coalition.org/>
- <http://www.steps-charity.org.uk/>

For emotional support there is also a worldwide charity called Befrienders Worldwide which you may find helpful: <http://www.befrienders.org>

Finally, I would like to thank you once more for taking part and I wish you all the very best for the future.

Kind regards,

James Oliver
Trainee Clinical Psychologist
Lancaster University

Appendix H: Expression of Interest Form

Research Study: Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

Name: _____

Gender: _____

Ethnicity: _____

Address: _____

Country of Residence _____

How old is your child? _____

Is your child male or female? _____

Child's ethnicity _____

How long has your child had available to use a prosthesis? Years _____ Months _____

Does your child have a health condition that may limit or preclude prosthesis use, or any other physical and/or intellectual disability? (please circle) **Yes/No**

I am interested in taking part in this study. Please contact me on:

Telephone number _____

Email _____

I would prefer to be interviewed via (please tick)

Skype _____ Telephone _____ Face-to-Face _____

Would you like a copy of the research paper or a summary when it is finalised?

Full Copy: **Yes/No**

Summary: **Yes/No**

Signed: _____

Date: _____

Applicant: James Oliver
Supervisor: Craig Murray
Department: Health Research
FHMREC Reference: FHMREC15092

12 July 2016

Dear James

Re: Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

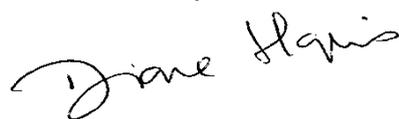
Thank you for submitting your research ethics application for the above project for review by the Faculty of Health and Medicine Research Ethics Committee (FHMREC). The application was recommended for approval by FHMREC, and on behalf of the Chair of the University Research Ethics Committee (UREC), I can confirm that approval has been granted for this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact the Diane Hopkins (01542 592838 fhmresearchsupport@lancaster.ac.uk) if you have any queries or require further information.

Yours sincerely,



Dr Diane Hopkins
Research Integrity and Governance Officer, Secretary to FHMREC

**Faculty of Health and Medicine Research Ethics Committee (FHMREC) Lancaster University
Application for Amendment to Previously Approved Research**

1. Name of applicant:
2. E-mail address and phone number of applicant:
3. Title of project:
4. FHMREC project reference number:
5. Date of original project approval as indicated on the official approval letter (month/year):

6. Please outline the requested amendment(s)
Note that where the amendment relates to a change of researcher, and the new researcher is a student, a full application must be made to FHMREC

7. Please explain your reason(s) for requesting the above amendment(s):

- 8.

Guidance:

- a) Resubmit your research ethics documents (**the entire version which received final approval, including all participant materials, your application form and research protocol**), with all additions highlighted in yellow, and any deletions simply 'struck through', so that it is possible to see what was there previously.
- b) This should be submitted as **a single PDF** to [Diane Hopkins](#). There is no need to resubmit the Governance Checklist

Applicant electronic signature:

Date

Student applicants: please tick to confirm that you have discussed this amendment application with your supervisor, and that they are happy for the application to proceed to ethical review

Project Supervisor name (if applicable):

Date application discussed

You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application

Applicant: James Oliver
Supervisor: Craig Murray
Department: Health Research
FHMREC Reference: FHMREC15132

15 August 2016

Dear James

Re: Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

Thank you for submitting your research ethics amendment application for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for this research project.

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Tel:- 01542 592838

Email:- fhmresearchsupport@lancaster.ac.uk

Yours sincerely,



Dr Diane Hopkins
Research Integrity and Governance Officer, Secretary to FHMREC.



**Faculty of Health and Medicine Research Ethics Committee (FHMREC) Lancaster University
Application for Amendment to Previously Approved Research**

1. Name of applicant:
2. E-mail address and phone number of applicant:
3. Title of project:
4. FHMREC project reference number:
5. Date of original project approval as indicated on the official approval letter (month/year):
6. Please outline the requested amendment(s)
Note that where the amendment relates to a change of researcher, and the new researcher is a student, a full application must be made to FHMREC
7. Please explain your reason(s) for requesting the above amendment(s):
- 8.

Guidance:

- a) Resubmit your research ethics documents (**the entire version which received final approval, including all participant materials, your application form and research protocol**), with all additions highlighted in yellow, and any deletions simply 'struck through', so that it is possible to see what was there previously.
- b) This should be submitted as **a single PDF** to [Diane Hopkins](#) There is no need to resubmit the Governance Checklist

Applicant electronic signature:

Date

Student applicants: please tick to confirm that you have discussed this amendment application with your supervisor, and that they are happy for the application to proceed to ethical review

Project Supervisor name (if applicable):

Date application discussed

You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application

Applicant: James Oliver
Supervisor: Craig Murray
Department: Health Research
FHMREC Reference: FHMREC15139

06 September 2016

Dear James

Re: Exploring the experiences of being a parent of a child with limb difference who has been provided with an artificial limb.

Thank you for submitting your research ethics amendment application (#2) for the above project for review by the **Faculty of Health and Medicine Research Ethics Committee (FHMREC)**. The application was recommended for approval by FHMREC, and on behalf of the Chair of the Committee, I can confirm that approval has been granted for this research project.

As principal investigator your responsibilities include:

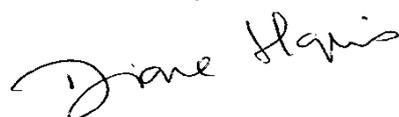
- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;
- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);
- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information.

Tel:- 01542 592838

Email:- fhmresearchsupport@lancaster.ac.uk

Yours sincerely,



Dr Diane Hopkins
Research Integrity and Governance Officer, Secretary to FHMREC.