Parenting a Child with Congenital Heart Defects

A thesis submitted to the University of Manchester for the degree of

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in the Faculty of Biology, Medicine and Health

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Thesis Abstract

Paper 1 is a metasynthesis and qualitative analysis of studies exploring coping in parents who have a child diagnosed with a congenital heart defect (CHD). Six databases were systematically searched using key words and index terms to identify the qualitative literature exploring parent coping strategies for managing the issues of having a child with CHD. Twenty-two studies met the inclusion criteria and quality appraisal conditions. These papers reported on 704 parents’ coping needs and strategies from their child’s diagnosis, undergoing multiple operations, providing health care and looking to the future. Four core themes emerged from the synthesis to describe the processes that enable parental coping: Emotional response, Support systems, Parental management and Avoidance. Recommendations were made for professionals who came into contact with families. Findings and limitations were discussed in relation to the existing literature.

Paper 2 explored the lived experience of 12 parents who had a child with single ventricle CHD, who had completed all stages of the Fontan surgical procedure. Interpretative Phenomenological Analysis (IPA) of the data identified three superordinate themes that captured the parent experience: Super-parenting, Accepting CHD and their role, and Striving for Normality. These themes were divided into further subthemes that explored the intricacies of the parenting journey from diagnosis, through surgeries and into childhood and adolescence. Clinical implications are discussed in relation to the role of psychologists working with families in health care settings.

Paper 3 is a critical reflection of the research. This paper discusses the rationale for the research design in both papers. Approaches to interviewing and analysis are reviewed with reference to researcher reflexivity, and a model of parent experience is proposed.
Declaration

No portion of the work referred to in the thesis has been submitted in support of an application for another degree or qualification of this or any other university or other institute of learning.
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Dedication

For my parents, who coped pretty well with our own dysfunctional lives.
Acknowledgements

The authors wish to thank the paediatric cardiology team at Alder Hey Children’s Hospital, particularly Lucinda Mawdsley for her invaluable assistance with recruitment, Dr Emma Twigg for stepping up to the role of field supervisor, and Mr Rafael Guerrero who recognised the need to learn from parents.

Thanks to my academic supervisors, Debbie Smith and Anja Wittkowski, for guiding me through this process. Additional thanks to Gokce Cokamay for screening papers and Katie Carpenter for quality checking papers for the systematic review.

My immense gratitude and respect goes to members of the ClinPsyD 2015 cohort, especially the ‘basement gremlins’, who supported each other, shared knowledge and provided much needed sustenance through the darkest days in our little basement room.

Finally, and most importantly, thanks to all of the parents who gave their time to openly tell their stories, in the hope that they would help new parents about to embark on their own rollercoaster journey.
Paper 1

Coping in Parents of Children with Congenital Heart Disease:

A Systematic Review and Meta-synthesis of the Qualitative Literature

Paper prepared for submission to Paediatric Cardiology

(See Appendix 1 for journal guidelines, this journal has no word limit for manuscripts)

Word count: 7684 (main text)

14,600 (complete text including tables, figures and references)
Abstract

Congenital heart defects (CHD) can be detected at ultrasound but are sometimes not diagnosed until birth, which can cause stress and heightened emotion within the family. Parents face challenges including dealing with surgical procedures for their child and integrating healthcare management into family life. The aim of this review was to understand parental coping with their child’s CHD.

Six databases were systematically searched to identify qualitative studies relating to parental coping in the context of having a child with CHD. Studies were subject to quality appraisal using Walsh and Downe’s (2006) checklist, and synthesised using Noblit and Hare’s (1988) meta-ethnographic approach, resulting in 22 studies reporting on 704 parents’ reports. The synthesis showed that parent coping fell within four overarching themes: Emotional Responses, Support Systems, Parental Management and Avoidance. These four themes contained a further 13 subordinate themes.

This is the first meta-synthesis of qualitative studies on parental coping with a child with CHD. Parental psychosocial coping varies over time from diagnosis, through surgery to childhood. Common themes are evident, but individuals employ their own styles and strategies based on prior experience, availability of social support, personal characteristics and beliefs. Parents try to maintain a sense of normality, integrating CHD into their lives without it having a major impact except at times of transition and hospitalisation when they must call upon additional strategies or supports to manage this stress.

Keywords: Family; adjustment; cope; child; CHD.
Introduction

Congenital Heart Disease (CHD) is one of the most common types of birth defects, affecting 8 per 1000 babies born globally [1], and prevalence has increased over time [2]. As there are many variations of CHD which sometimes occur in combination and with differing severities, prognosis and treatment may vary between individuals. NICE guidance [3] advises that “children, young people and their parents or carers may need support, and sometimes expert psychological intervention, to help with distress, coping, and building resilience” (p.14) and emotional and psychological wellbeing should be discussed with families regularly, particularly at times of transition. Chronic illnesses, such as CHD, and their associated complications can adversely impact family life [4-7]. Presence of CHD has been shown to increase the vulnerability of the whole family to psychological and social distress [8-10]. However, using the perspective of coping, many studies also show that parents and families manage to adjust to the presence and demands of childhood conditions [11,12]. For example, families can be put under strain of a chronic illness at various time points [13] including at diagnosis [14], birth [15], surgery [16] and adapting to the integration of healthcare alongside typical parenting [17,18].

The experiences of parenting a child with a chronic illness, and the impact of paediatric illness on families, have been reviewed [19,20]. In their review of 94 studies to explore the impact of CHD on families, Wei, Roscigno, Hanson and Swanson [21] identified major themes based on quantitative checklists of parents’ psychological health, family life, parenting challenges and family-focussed interventions. However, most of these studies were quantitative and used self-report measures with pre-defined questions and answers. As such, these studies could not have reported on the detailed influences of CHD on parents and certainly not in the parents’ own words. Furthermore, deductive methods use prior
assumptions that may not answer the research question [22]. In contrast to quantitative methods, qualitative methods provide details about human behaviour, emotion and personality characteristics and they enable researchers to make sense of patterns in the meaning through differences and divergence, particularly in health research [23].

In another review of 25 studies, featuring mostly quantitative designs but also including some qualitative papers, Jackson, Frydenberg, Liang, Higgins and Murphy [24] identified that the impacts on a family can include psychological distress and wellbeing, family functioning and quality of life. Coping styles varied dependent on age and gender; mothers sought social support, “vented” about their situation and turned to spirituality or religion, whereas fathers were more likely to use alcohol as a coping mechanism.

Despite qualitative studies informing the evidence base by providing rich, in depth details of participant experiences [25], to date, no review has solely focussed on the lived experiences of parents of children with CHD and their self-reported coping. Therefore, the aim of this meta-synthesis was to focus on the qualitative reports of parents in order to develop a richer understanding of how parents cope with a child with CHD. Another aim was for this synthesis to further the understanding of the social, emotional and practical coping mechanisms that parents engage with when they have a child diagnosed with CHD.

Coping is a broad term, and so various definitions were considered [26,27] including those specific to psychological adjustment to chronic illness [28,29] which had been used in other studies on parental coping [30,31]. These definitions shared similarities relating to the problem-focus or emotion-focus of distress, and the subsequent responses intended to reduce the burden of stressful life events. The Snyder [27] definition of coping refers to any strategies that effectively manage emotional, physical or psychological burdens, and was
considered sufficient and appropriate to cover the breadth of literature exploring parental coping.

We set out to answer the following research question: How do expectant parents and parents of children with CHD cope with their experience from diagnosis, surgery and beyond?

Methodology

The meta-ethnographic approach described by Noblit and Hare [32], frequently used in health care research [33], was selected to synthesise the findings from the included studies. It is a commonly used idealist technique to synthesise and explore differences between studies [34], whilst preserving the rigour and quality of the primary data [35].

The main themes and concepts identified from systematic searches of the qualitative literature were synthesised to establish broader themes across the studies to be included in this review. The seven-step-approach [32] was chosen to guide the synthesis process (see Appendix 2) which consists of: Research Question; Systematic Review Process; Reading and rereading studies: Identify primary and secondary themes; Determine how studies are related to each other; Translate the studies into one another; Synthesise translations; Express the synthesis.

Search strategy

A systematic search of six databases was conducted in October 2017 (MEDLINE, CINAHL, PsycINFO, PubMed, ProQuest and Web of Science). Online databases were systematically searched by the first author (ML). Keywords from four broad areas relating to CHD, coping,
family and child (see Table 1 for further details) were truncated. Multiple synonyms of search terms were utilised using Boolean search operators AND/OR across all databases, with exploded or Medical Subject Heading (MeSH) terms, when available (see Appendix 4). Broad search terms were selected to cover a wide range of papers based on the SPIDER search tool [36].

Hand searching was also undertaken. First, the reference lists of identified papers were scrutinised for additional papers. Secondly, Google Scholar was searched for additional studies using simple broad-based terms to identify any remaining studies [37].

Table 1: Search terms used in systematic literature search

<table>
<thead>
<tr>
<th>Domain</th>
<th>Search terms</th>
</tr>
</thead>
<tbody>
<tr>
<td>S</td>
<td>(Family OR parent* OR mother OR father OR mum OR dad OR carer OR caregiver OR famili* OR maternal OR paternal) AND (child* OR adolescen* OR p<em>ediatric OR infant</em> OR f*etus OR offspring OR son OR daughter OR baby OR babies OR newborn OR toddler)</td>
</tr>
<tr>
<td>PI</td>
<td>Congenital heart disease* OR congenital heart defect OR heart defect* OR single ventricle</td>
</tr>
<tr>
<td>D</td>
<td>*Design type was not specified as we wanted to include all qualitative research designs</td>
</tr>
<tr>
<td>E</td>
<td>Adjust* OR adapt* OR cope OR coping OR manage* OR deal OR endure OR withstand</td>
</tr>
<tr>
<td>R</td>
<td>Qualitative</td>
</tr>
</tbody>
</table>

Inclusion and exclusion criteria

Studies were included in the review based on the following eligibility criteria: 1) an empirical study collecting qualitative data published in peer reviewed journals, 2) reporting on experiences of parents of infants and children diagnosed with congenital heart disease including how they coped at various stages, 3) included studies utilised samples of parents,
over 18 years of age, who had a child who had been diagnosed with a congenital heart defect (CHD). The Snyder [27] definition of coping was used in our selection of studies so that any paper which made reference to parents managing an emotional, physical or psychological burden was included. No restriction was placed on language or year of publication. Studies deemed as very poor quality (see Quality Appraisal below for details) were excluded from the review.

Management of search outcomes and eligibility screening

The search process is illustrated in Figure 1. The researchers adhered to PRISMA guidelines [38] for systematic reviews and metasyntheses and registered the review on PROSPERO (CRD42017049683) prior to searching [39]. The first author screened the search results against the identified eligibility criteria. An independent researcher screened 10% of the search results to provide a measure of reliability of the screening process. The agreement between researchers was 100%.
Figure 1. PRISMA flow diagram outlining process of study identification

Records identified through database searching (n = 674) [Medline (n = 21); CINAHL (n = 23); PsycINFO (n = 56); PubMed (n = 373); ProQuest (n = 90); Web of Science (n=111)]

Records after duplicates removed (n = 477)

Records excluded (n = 197)

Records excluded (n = 425)
  Reasons:
  Topic N/A (n=134)
  Not parents (n=32)
  Not CHD (n=241)
  Not coping (n=12)
  Publication type (n=6)

Records screened by title/abstract (n = 52)

Additional records identified through other sources (n = 12)
  [Ref lists (n= 4), Google Scholar (n = 8)]

Records excluded (n = 37)
  Reasons:
  Not parents (n=5)
  Not CHD (n=14)
  Not coping (n=13)
  Publication type (n=5)

Full-text articles assessed for eligibility (n = 15)

Records excluded (n = 425)
  Reasons:
  Topic N/A (n=134)
  Not parents (n=32)
  Not CHD (n=241)
  Not coping (n=12)
  Publication type (n=6)

Studies included in qualitative synthesis (n = 22)

Papers removed based on poor quality (n = 5)

Papers screened for quality (n = 27)

Eligibility

Quality

Included

Screening

Identification
Quality appraisal

The Walsh and Downe [40] quality appraisal checklist (see Appendix 3 for a copy) was used to assess 12 different aspects of methodological and interpretive rigour in the included qualitative studies. In contrast to the 10-item-CASP [41], this checklist offers a more comprehensive framework with clear descriptions for each criterion.

This checklist contains 12 criteria covering eight stages of research from scope and purpose to relevance and implication of findings. Studies were rated against each of the 12 items and awarded a point if they met criteria fully. As it is common for studies to meet some but not all of the points within a criterion, it was decided to award 0, 0.5 or 1 point, producing a maximum score of 12. Although a total score can be calculated, it should only be seen as a guide to the overall methodological quality of each paper [42]. Thus, the following categories were used: Category A represented studies which were rated as high quality and low methodological bias; these studies scored between 9 and 12. Category B studies were rated as moderate quality; these studies scored between 6 and 8. Category C studies scored below 6 (less than half of the maximum score) and were considered to represent low quality and high methodological bias; therefore, they were excluded from this meta-synthesis [43].

The methodological quality of the identified studies was appraised by the first author (ML). Thirty per cent of papers were reviewed by an independent researcher (KC) to verify the accuracy of the quality assessment.
Results

Study characteristics

The literature search yielded 22 studies that reported on parental coping with a child diagnosed with a CHD, or a prenatal diagnosis. The characteristics for these studies can be seen in Table 2. Studies were from the USA (n=10), Canada (n=6), Australia (n=2), UK (n=1), Japan (n=1), Sweden (n=1) and Taiwan (n=1). The methods of analysis used in the studies included content analysis (n=7), grounded theory (n=5), phenomenological analysis (n=3), thematic analysis (n=3) and framework analysis (n=1). Three studies used secondary analysis and did not detail a specific methodology. Most studies used purposive or convenience samples from health centres and hospitals associated with the researchers. The vast majority of studies used interviews to collect data, with three reusing data sets through secondary analysis of existing interview transcripts and memos. Two studies used focus groups, while one examined journal entries and one asked open-ended questions as part of a survey. The median sample size was 16, ranging from 8 to 175 participants.

The studies represented data from 704 parents (511 mothers, 176 fathers, 17 unreported), aged between 18 and 60 years, of children pre-birth to 19 years old. Ethnicity and socioeconomic status varied within and between studies. Children’s diagnoses covered a broad range of defects classed as CHD including atrial septal defects, ventricular septal defects, tetralogy of fallot and hypoplastic left heart syndrome (see “Child characteristics” in Table 2 for details). Some studies focused only on the experience of pregnancy, while others explored parental coping throughout their child’s life. Details of the final 22 studies are summarised in Table 2.
<table>
<thead>
<tr>
<th>Reference and country</th>
<th>Research aims</th>
<th>Participant characteristics</th>
<th>Child characteristics</th>
<th>Data collection method / Method of analysis</th>
<th>Main findings/themes</th>
</tr>
</thead>
<tbody>
<tr>
<td>1 Wei, Roscigno and Swanson [44] USA</td>
<td>To describe parents’ perceptions of healthcare providers’ actions when their child is diagnosed with CHD and undergoes heart surgery</td>
<td>10 mothers 3 fathers Age: &lt;20=1, 20-29=6, 30-39=5, 40-49=1</td>
<td>10 children (9 boys, 1 girl) &lt;1 y.o = 7, 1-2 y.o = 2, 5 y.o=1 Atrial septal defect =1 Ventricular septal defect =4 Atrial ventricular septal defect =1 Tetralogy of Fallot =3 Pulmonary stenosis =1</td>
<td>Interviews/ Directed content analysis</td>
<td>Healthcare providers play an imperative role in helping parents understand and adjust to their child's ongoing medical conditions, the effective ways of doing which are to include them in their child's treatment and validated their roles as parents by incorporating them in their child’s daily hospital care.</td>
</tr>
<tr>
<td>2 Nakazuru, Sato and Nakamura [45] Japan</td>
<td>To examine the stress and coping of mothers whose infants needed CHD surgery and to identify the factors that influenced maternal coping</td>
<td>11 mothers aged 30-41, (mean = 36)</td>
<td>5 boys, 6 girls ≥ 1 month, ≤ 6 years Atrial septal defect =3 Atrial ventricular septal defect =5 Tetralogy of Fallot =1 Atrial-ventricular block=1 Double outlet right ventricle=1</td>
<td>Semi-structured interviews/ Qualitative content analysis</td>
<td>5 factors that influenced Japanese mothers' coping: the diagnosis event, the number of symptoms of CHD, the anticipated number of surgeries, the presence of chromosomal abnormalities, and the infant's age. And further, our subjects, Japanese mothers, reported that they held back emotions and evidence of their anxiety. The coping of mothers began when they first received the diagnosis and changed as the surgery date approached. Japanese mothers tried to prepare for surgery without a reliable schedule for surgery and without enough educational preparation. They embraced their high levels of anxiety but dealt with surgery by trusting the health care team.</td>
</tr>
<tr>
<td>3 Golfenshtein, Deatrick,</td>
<td>(1) To investigate coping mechanisms</td>
<td>14 mothers</td>
<td>14 children (11 boys 3 girls)</td>
<td>Focus groups/ Qualitative content</td>
<td>Mothers use both passive and active coping mechanisms deal with stress. The active coping</td>
</tr>
<tr>
<td><strong>Lisanti and Medoff-Cooper [46] USA</strong></td>
<td>of mothers whose infant with complex CHD is admitted in the CICU, and (2) to explore the acceptability and feasibility of mindfulness as a potential stress-reduction intervention for these mothers.</td>
<td>11-63 days old (median=19) Single ventricle=10 Bi-ventricle=4</td>
<td>analysis</td>
<td>strategies include seeking social support, focusing and doing things for their baby, and positively repring optimistic scenarios. The passive strategies include denial and distraction, which were used by all mothers. Most mothers also use emotion regulation focused coping, rather than problem solving focused coping, as they feel that there are only few things they could do for their baby in the CICU setting.</td>
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<tr>
<td><strong>McKechnie, Pridham and Tluczek [47] USA</strong></td>
<td>The twofold purpose of this study was to (a) explore and qualitatively describe parents’ caregiving motivation to manage maternal–fetal and infant health care and (b) examine potential links between parents’ motivation to manage health care and their symptoms of distress subsequent to a fetal diagnosis and after-birth treatment of the infant’s CCHD.</td>
<td>6 mothers aged 23-34 (mean=30) 6 fathers aged 24-35 (mean=31)</td>
<td>6 children Hypoplastic left heart, coarctation of aorta, transposition of the great arteries, and tetrology of Fallot</td>
<td>Semi-structured interviews / Content Analysis</td>
<td>Based on parents’ descriptions, three categories of the motivation to manage health care were identified and included efforts (a) to determine expectations of health care providers, (b) to reconcile illness- and non-illness-related care, and (c) to express agency as a parent. Although variations in this motivation were found among parents over time, the same three categories were identified in parents’ narratives during pregnancy with a fetal diagnosis and after birth with an infant requiring intervention.</td>
</tr>
<tr>
<td><strong>Meakins, Ray,</strong></td>
<td>The purpose of this exploratory study</td>
<td>17 fathers, aged 27-50 12 boys, 12 girls Children’s ages ranged</td>
<td>Data from 2 original grounded theory studies</td>
<td>Parental response to caring for a child with HLHS was characterized by both vigilant and</td>
<td></td>
</tr>
</tbody>
</table>
Hegadoren, Rogers and Rempel [48] Canada

was to address the following research question: Is the parenting process among parents of children with HLHS characterized by exaggerated vigilant parental action, and if so, how does this influence parental response? The intent was to differentiate the varying intensities of vigilant parental action.

(24) mothers aged 22-48 (mean=32.8) from 2-60 months (mean=27.9 months) including 55 interviews from 41 parents/Directed secondary thematic content analysis

Health professionals taught parents to vigilantly attend to their baby’s complex care requirements, which involved skills such as feeding, weighing, administering medications and oxygen; monitoring all aspects of the baby’s health; and taking action based on subtle changes. There was evidence that vigilant parental actions were exaggerated at times, with parents focusing on the child’s care and monitoring at the expense of other aspects of their life.

McKechnie, Pridham and Tluczek [49] USA

The purpose of the study reported here was to address this gap in the theoretical and empirical literature by examining how parenting develops after a major fetal anomaly diagnosis.

25 pregnant women aged 18-46 (mean=28) 12 men aged 19-52 (mean=30) 15 female foetus, 9 male, 2 unknown. Fetal diagnoses included hypoplastic left and right heart syndromes, tetralogy of Fallot, double outlet right ventricle, double inlet left ventricle, coarctation of the aorta.

Interviews/Grounded dimensional analysis

Findings revealed that preparing heart and mind was a dynamic and nonlinear process. Fetal and future child health information, experiences of loss, and social interactions influenced the preparing process. These influencing conditions triggered turning points, described as realizing a different perspective. Expectant parents explained that turning points led them to develop, amend, and anticipate certain strategies for preparing emotionally and cognitively for becoming parents. These strategies included efforts to proceed in service of the child, to address external needs, and to integrate experiences with the normative approach to pregnancy.

Kosta, Harms, 63 fathers, (mean age = 35) 43 boys, 48 girls. Coarctation of the aorta.

Telephone or face to face interviews/

Parents reported a range of difficulties from dealing with their baby’s unfolding illness,
<table>
<thead>
<tr>
<th>Study Authors</th>
<th>Population</th>
<th>Methodology</th>
<th>Data Collection</th>
<th>Analysis</th>
<th>Findings</th>
</tr>
</thead>
<tbody>
<tr>
<td>Franich-Ray, Anderson, Northam, Cochrane, Menahem and Jordan [50] Australia</td>
<td>Parental experiences of their infant’s hospitalization for cardiac surgery and to identify factors that may be targeted in the future to alleviate sources of parental distress.</td>
<td>Thematic analysis</td>
<td>Open ended questions on a survey/Content analysis</td>
<td>Surgical and recovery to the structural and systemic issues associated with the broader context of their experiences. In an effort to be near their baby, parents struggled to obtain the necessities for daily living at the hospital and negotiate transitions between their hospital and home lives throughout the admission. Domains parents identified for change included the availability of facilities and resources and the quantity and quality of information and emotional support. Parents reported relationships with hospital staff as the most common source of support during this challenging time.</td>
<td></td>
</tr>
<tr>
<td>Sira, Desai, Sullivan and Hannon [51] USA</td>
<td>The purpose of this study was to: a) identify existing coping strategies in mothers who have a child diagnosed with CHD based on McCubbin et al.’s (1983) three coping patterns (I, maintaining family integration and optimism; II, maintaining self-esteem; and III, maintaining psychological stability). b) Identify the most used coping pattern among study participants, while maintaining self-esteem and psychological stability was the least utilized pattern.</td>
<td>Open ended questions on a survey/Content analysis</td>
<td>Understanding the medical situation and communication with other parents was the most used coping pattern among study participants, while maintaining self-esteem and psychological stability was the least utilized pattern.</td>
<td></td>
<td></td>
</tr>
<tr>
<td>Study</td>
<td>Authors</td>
<td>Purpose</td>
<td>Participants</td>
<td>Methods</td>
<td>Findings</td>
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</tr>
<tr>
<td>9</td>
<td>Bruce, Lilja and Sundin [52] Sweden</td>
<td>The purpose of this study was to illuminate the meanings of support as disclosed by mothers of children with congenital heart defects.</td>
<td>10 mothers Mean age 38 years, 10 Children aged 3-12 years</td>
<td>Narrative interviews/Phenomenological-hermeneutic method</td>
<td>The comprehensive understanding of mothers’ lived experiences of support emerged as the experiences of receiving good support, receiving “poor support,” and absence of support.</td>
</tr>
<tr>
<td>10</td>
<td>Rempel, Ravindran, Rogers and</td>
<td>Our aim was to generate evidence to inform clinical</td>
<td>15 mothers 10 fathers 17, 15 children Aged 6 months-4.5 years</td>
<td>Interviews/Constructivist grounded theory</td>
<td>A process of Parenting under Pressure emerged that was characterized by four overlapping and re-emerging phases: (1) realizing and adjusting</td>
</tr>
<tr>
<td>Reference</td>
<td>Country</td>
<td>Study Design</td>
<td>Sample Characteristics</td>
<td>Data Collection Methods</td>
<td>Data Analysis</td>
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</tr>
<tr>
<td>Magill-Evans [53] Canada</td>
<td>Canada</td>
<td>Qualitative descriptive study</td>
<td>Practice with parents of young children with HLHS based on the perspectives of both parents and grandparents using a recent treatment cohort. The research question was: What is the process of parenting young children with HLHS from the time of diagnosis through the survival of the first two surgeries and survival or anticipation of the third surgery?</td>
<td>Journal entries and survey by email/Colaizzi’s steps to analysis of phenomenological data</td>
<td>Data analysis of the journal entries revealed six major themes during the time period of the days before surgery, the day of surgery and days after surgery: 1) Feeling Intense Fluctuating Emotion, 2) Navigating the Medical World, 3) Dealing with the Unknown, 4) Facing the Possibility of My Baby Dying, 5) Finding Meaning and Spiritual Connection, and 6) Mothering Through It All. These themes frequently occurred together and were woven throughout the descriptions of the mother’s experiences.</td>
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<tr>
<td>Harvey, Kovalesky, Woods and Loan [54] USA</td>
<td>USA</td>
<td>Qualitative descriptive study</td>
<td>The purpose of this qualitative descriptive study was to explore the lived and recalled experiences of mothers of infants with moderate to severe CHD who were diagnosed post-natally and needed heart surgery prior to their first birthday.</td>
<td>Journal entries and survey by email/Colaizzi’s steps to analysis of phenomenological data</td>
<td>Data analysis of the journal entries revealed six major themes during the time period of the days before surgery, the day of surgery and days after surgery: 1) Feeling Intense Fluctuating Emotion, 2) Navigating the Medical World, 3) Dealing with the Unknown, 4) Facing the Possibility of My Baby Dying, 5) Finding Meaning and Spiritual Connection, and 6) Mothering Through It All. These themes frequently occurred together and were woven throughout the descriptions of the mother’s experiences.</td>
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<tr>
<td>Number</td>
<td>Author(s) and Location</td>
<td>Study Title</td>
<td>Participants</td>
<td>Methodology</td>
<td>Findings</td>
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<td>12</td>
<td>Cantwell-Bartl and Tibballs [55] Australia</td>
<td>To evaluate the psychosocial status of mothers and fathers of infants with hypoplastic left heart syndrome while in the PICU.</td>
<td>16 mothers 13 fathers 16 children aged 1-19</td>
<td>Semi-structured face to face narrative interviews/Thematic analysis</td>
<td>All parents reported multiple stresses which commenced with their infant’s diagnosis and endured throughout their infant’s time in PICU. Of 11 parents whose infants were diagnosed with hypoplastic left heart syndrome postbirth, six of seven mothers had acute stress disorder and one mother had posttraumatic stress disorder, and of four fathers, two fathers had acute stress disorder and one father had posttraumatic stress disorder. Parents also experienced losses. Many parents were marginalized from their infant’s care by the environment of PICU. Fifty percent of mothers experienced difficulties with parental-infant bonding. Ten parents (34%) began the process of adaptation to their infant’s hypoplastic left heart syndrome and were assisted by the support and sensitivity of staff or had discovered other resources.</td>
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<tr>
<td>13</td>
<td>Rempel, Rogers, Ravindran and Magill-Evans [56] Canada</td>
<td>The purpose of this study was to conceptualize the needs of parents of young children with hypoplastic left heart syndrome (HLHS) to provide a theoretical framework to inform the development of future parent interventions. Specific research questions included</td>
<td>15 mothers 10 fathers 15 children 6 months to 4.5 years all with hypoplastic left heart syndrome</td>
<td>53 previously conducted interviews, and conceptual memos written by the researchers/Secondary analysis of grounded theory data</td>
<td>Persistent stress and uncertainty characterized parents’ and grandparents’ accounts. Parents understood the life-threatening nature of HLHS by knowing a child who died or by their own child nearly dying. Parents hoped for the best while simultaneously acknowledging the worst-case scenario. Despite their child’s uncertain future, many parents felt lucky or grateful, especially in comparison to others, and described their personal growth through parenting a child with HLHS. Grandparents’ “birds-eye” view of their adult child’s experiences corroborated parents’ accounts and provided additional insights.</td>
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</table>
(1) what are the common experiences and needs of parents of young children with HLHS? (2) How can these experiences and needs be synthesized to optimize translation of evidence for health care professionals? and (3) How can the resulting conceptualization inform future interventions?

<p>| 14 | Rempel, Blythe, Rogers and Ravindran [57] Canada | Research Question 1: What is the process of family management related to the dimensions of the FMSF in families of infants with HLHS who underwent the early era Norwood procedure in comparison to parents of children who underwent the later era Sano procedure? Research 24 mothers 17 fathers. Mean age=33.7 12 boys: mean age=23.2 months 12 girls: mean age=32.8 months 55 previously conducted interviews/ Secondary thematic content analysis | The process of family management that emerged in this study began with the diagnosis of the baby’s HLHS, included the baby’s survival of the first surgery, and continued until going home with baby. Each of these time periods was characterized by one family management style more than the others and differences between the early era (Norwood) and late era (Sano) cohorts were evident when particular dimensions of the family management styles were considered. The key finding of this study was that parents demonstrated an intense, dynamic, and transforming process of family management from the diagnosis stage through to the baby |</p>
<table>
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<th>Question 2: How does family management change over time from the initial diagnosis through the early period of home care?</th>
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<td>15</td>
<td>The purpose of this theoretically guided qualitative inquiry was to examine parents’ experiences in terms of prebirth caregiving motivations following a prenatal fetal CCHD diagnosis reported retrospectively 1 to 3 months following birth.</td>
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</table>
|   | 13 mothers, 3 fathers  
Aged 21-39 years  
14 children. All had multiple diagnoses, including hypoplastic left heart syndrome, tetrology of Fallot, and other serious aortic and valve defects.  
Semi structured interviews/Directed content analysis.  
The prenatal diagnosis impacted all aspects of the caregiving system. The motivation to become a caregiver appeared to activate the caregiving system as a whole, to reach the primary goal of protecting, nurturing, and comforting the offspring (Bowlby, 1988). Beginning before birth and continuing throughout the first year of life, parents sought to protect, nurture, and comfort their offspring in various ways. However, after receiving a prenatal diagnosis of a CCHD, parents in this sample vividly recalled directing significant energy toward preparing heart and mind, which involved their varied emotions, thoughts, and states of being. |
| 16 | To examine the process of parenting a child with hypoplastic left heart syndrome who had survived the Norwood surgical approach. The specific research question is “How do 9 mothers, aged 22-48 mean 34 years  
7 fathers, aged 30-50, mean 36 years  
9 children: 3 boys, 6 girls aged 2 months to 5 years  
All with hypoplastic left heart syndrome  
Interviews/Constructivist grounded theory  
Parents used normalization in the context of uncertainty regarding the ongoing survival of their child. Parents described their underweight children as being on their own growth curve, and viewed their developmental progress, however delayed, as reason for celebration, as they had been prepared for their child to die. |
<p>| 15 |   |   | surviving the first of three surgeries and going home. The parents whose babies were treated in the era that had improved survival rates had a more positive view of their child’s illness and therefore showed more thriving family characteristics than the parents whose babies were in the early treatment era. |</p>
<table>
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<tr>
<th>Source</th>
<th>Research Question</th>
<th>Methodology</th>
<th>Participants</th>
<th>Data Collection</th>
<th>Findings</th>
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<tbody>
<tr>
<td>17 Rempel and Harrison [60] Canada</td>
<td>To describe the parenting experience of mothers and fathers whose child with HLHS underwent treatment that included a series of high-risk surgeries starting with the Norwood surgical procedure soon after birth.</td>
<td>9 mothers, aged 22-48 mean 34 years 7 fathers, aged 30-50, mean 36 years</td>
<td>9 children: 3 boys, 6 girls aged 2 months to 5 years All with hypoplastic left heart syndrome</td>
<td>Unstructured interactive interviews, face to face and phone based/ Constructivist grounded theory</td>
<td>The mothers and fathers in this study exemplified extraordinary parenting. Their children with HLHS were survivors of advancing surgical technology and a learning experience for many health care professionals, who therefore were not in a position to provide parents with direction for care.</td>
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<tr>
<td>18 Lan, Mu and Hsieh [61] Taiwan</td>
<td>To investigate the essence of the experience of mothers during the decision-making process when facing their less than three-year-old child undergoing heart surgery due to 6 tetralogy of fallot, 3 ventricular septal defect (1 also transposition of the great arteries and atrial septal defect)</td>
<td>9 mothers Aged 27-39, mean 32 years</td>
<td>9 children: 5 female, 4 male</td>
<td>Interviews/ Colaizzi’s Phenomenological Analysis</td>
<td>The essence of the maternal experience themes during the decision-making process included (i) understanding the surgery step by step, (ii) role pressure, (iii) constructing care-taking ability, (iv) endeavouring to maintain family functioning while preparing for surgery and (v) deliberate consideration to make the correct decision.</td>
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<tr>
<td>Study</td>
<td>Authors</td>
<td>Methodology</td>
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<td>19</td>
<td>Leuthner, Bolger, Frommelt and Nelson [62] USA</td>
<td>To examine the impact of an abnormal fetal echocardiogram (echo) on expectant parents’ experience of pregnancy. We hypothesized that the fetal echo marked a critical point of emotional response, coping and decision making, and that we would observe differences in the way men and women respond to the information and experience provided by this technology.</td>
<td>9 mothers, 7 fathers. Aged 28-36 years. 11 pregnancies at 16-34 weeks gestation at time of fetal ECG. 2 complex single ventricle, congenital heart block and cardiomyopathy, transposition of the great arteries, truncus arteriosus, TAPVR, Epstein’s anomaly, Hypoplastic left heart syndrome.</td>
<td>Focus groups / Thematic content analysis</td>
<td>The women expressed strong emotions of guilt, fear and sadness or hopelessness. They coped by attaching and bonding to the fetus, and were realistic about the future. The men experienced emotions of anger and anxiety. They coped by remaining optimistic, and focused on supporting their wives. Men grieved after the birth of the baby. The fetal echo was felt to be beneficial and not harmful by the parents. The fetal echo led to significant changes in the couples’ perceptions of themselves and their relationships. Clinicians performing fetal imaging must be aware of parental affective responses and coping mechanisms in order to provide the required psychological support.</td>
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<td>20</td>
<td>Kendall, Sloper, Lewin and Parsons [63] UK</td>
<td>To obtain the views of their parents about the need for, and shape of, services for rehabilitation.</td>
<td>17 parents, 19 children aged 5-18</td>
<td>Interviews / Framework analysis</td>
<td>Parents would welcome more help and support from health professionals to enable them to manage more effectively the condition with their children. Particular areas of concern relate to the information they receive about the condition; communication between themselves and health professionals; establishing safe levels of activity; and managing the condition at school.</td>
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</table>
| 21 | Gantt [64] USA | To study the effect of congenital heart disease (CHD), a | 11 mothers, 11 daughters | Unstructured interviews / Content analysis | Normalizing in the face of chronic illness was the overriding theme of the study. Two subthemes related to normalizing arose from the data. “It’s
potentially life-threatening chronic illness, on the mother–daughter relationship. “No big deal” was found to best describe how most mothers and their daughters with heart disease viewed their lives and relationships. One other subtheme, “Sometimes it’s a very big deal,” was described by those mothers and daughters who could not, due to ongoing health problems, normalize their lives. Relationships with family as well as age, severity of illness, and developmental stage, were seen as mediators of normalizing in the lives of the participants.

| 22 | Gudmundsdottir, Gilliss, Sparacino, Tong, Messias and Foote [65] USA | Exploratory study, the broad purpose of which was to examine the impact of CHD on the parent-adolescent dyad during adolescence. | 7 mothers 1 father | 8 children (3 girls, 5 boys) Age at interview 13-25 | Semi-structured interviews / Grounded theory | There were two distinct patterns of coping within the parent-adolescent dyad: (1) coping that was similar between parent and child, and (2) coping that was dissimilar. Within each of these categories were subcategories that referred to the styles of managing the illness situation. The category of similar coping included the subcategories of (a) mutual protection; (b) being accepting of each other; (c) dealing with the illness by normalizing it; (d) approaching the illness mechanically; and (e) keeping the illness in its place. A parallel problematizing and withdrawing mode of managing the illness was the only subcategory identified under dissimilar coping. | aStudies using the same sample
Quality assessment of methodology

All of the eligible studies were screened against Walsh and Downe [40] criteria. Table 3 provides an overview of scores for each included study. Eight of the studies included in this meta-synthesis were rated as category A and 15 were rated as category B, of which four papers [48,50,57,61] were borderline to category A. Five papers were excluded due to poor methodological quality and lack of rigour and detail [63,66-70] (see Appendix 5 for the quality assessment of excluded papers).

As can be seen in Table 3, the majority of studies did not report fully on all areas of methodology as described in the checklist\(^1\). Only two of the 22 studies explicitly referred to researcher reflexivity within the research process [52,56], which is considered a general weakness of the included studies.

The rating and categorisation process of 30% of the initial 27 studies was also checked by an independent researcher for reliability. There was 75% agreement in the categorisation of the ratings. Both researchers agreed to exclude one paper that did not meet quality rating standards for inclusion in this review.

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\(^1\) For more detail on quality appraisal, see critical evaluation paper.
Table 3: Quality assessment of reviewed studies

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<th>Criteria</th>
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<td>1: Clear statement of, and rationale for, research question/aims/purposes</td>
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<td>3: Method/design apparent, and consistent with research intent</td>
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<td>11: Demonstration of sensitivity to ethical concerns</td>
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A = Excellent, B = Good.
Synthesis

Despite differences between the studies in terms of aims, methodology and analysis, there were similarities in the findings, suggesting a shared sense of reported parent coping with a child with a diagnosis of CHD. Four overarching themes emerged from the synthesis, which contained 13 sub-ordinate themes. Figure 2 illustrates the structure of themes and sub-themes, while Table 4 shows whether they were present, refuted, or absent in the 22 studies.
Figure 2: Representation of synthesised themes and subthemes and their presence during diagnosis, surgery and childhood stages

Present at diagnosis, surgery and throughout childhood

Theme 1
Emotional Response

Express/withhold emotion
Attachment or detachment
Positive Thinking

Theme 2
Support Systems

Within couple
Peers/Family
Professionals
Faith/Spirituality

Theme 3
Parental Management

Striving for normality
Becoming experts
Hypervigilance

Theme 4
Avoidance

It’s out of my hands
Privacy
Denial and distraction
Theme 1: Emotional responses

Most of the studies detailed parents’ emotional responses as part of their coping, at the initial discovery of their child’s diagnosis as well as during the surgeries. This theme comprised of three further subthemes that expanded on the category.

Subtheme 1: Expressing or withholding emotion

This theme describes the expression of emotion in relation to their child, but also considers parents’ use of suppression or denial in order to protect themselves from overwhelming emotions, such as shock, grief, guilt, hopelessness, anxiety, anger, sadness and fear: “To define in words the emotion, questions, uncertainty, emptiness, and broken heart that I carried with me through the situation is impossible” [54]. Participants in 11 out of 22 studies talked about experiencing extremes of emotion and expressing these: “When my son was in surgery, I cried uncontrollably. I wanted someone to be with me, give me a little hand-holding, and tell me he’s going to be ok.” [44], showing how the expression of emotion functioned to communicate to others the need for support. One parent described the strong anger they felt after hearing the diagnosis which has prevailed throughout their child’s life: “I had a lot of anger at God. I was furious and I still am” [62].

In contrast, and only reported in a few studies, some parents described suppressing their emotions in order to cope with their everyday lives: “I am very emotional but I cannot keep walking around with sad face, be depressed. If I get sick who is going to take care of my kids?” [46].
Subtheme 2: Attachment or detachment

This theme captured how parents bonded with their child both before s/he was born and just after birth, prior to their first surgery. These were precarious periods when parents did not know the extent of their child’s complications, nor whether they would survive through a complex operation at such a young age.

Parents who treated the foetus as a child before they were even born felt as though s/he was more real, and they were able to form loving attachments even in the face of possible loss: “It’s your kid [child]. You’re the parent of that kid [in utero] that has the birth defect. And it’s just such a hard thing to just do, day by day and everything. This is why I’m feeling this way, because I am the parent” [49]. “It’s my baby and I love him immensely. I loved him before he was born and I loved him when he was born” [58].

Other parents did not form the same attachments and reported feeling detached from their child before they were born because they did not know if they would survive: “We didn’t know if she would make it through the first surgery or anything.... not to say you’re detached but you’re a bit guarded... I loved her and I knew that we wanted to keep her but you don’t connect” [53]. During the surgery when there was a real risk of losing their child, parents’ fear of forming attachments made them avoid getting too close: “I guess in the back of my mind, I felt that, if she was going to die, then probably the sooner the better because I just knew with time we’d get more and more attached to her” [57] and some did not want to even think about growing close to their child until they knew it would survive: “take him away, make him better and then I . . . may be able to bond” [62].
Subtheme 3: Positive Thinking

This theme explored how parents coped with the often honest or blunt prognosis of their child’s condition. Cardiologists typically informed them of the odds that were not necessarily in their favour, and parents were given the decision of whether to terminate or proceed with pregnancy, then give their child either palliative care or the process of open heart surgery. Parents coped with these dilemmas by believing their child was on the other side of the odds, hoping that they were not part of the risk statistic and doctors were wrong with their prognosis: “Hope! Yeah, that’s all we had, really. I think that throughout the whole thing, the only hope that we ever had was basically that they [diagnosing physicians] were wrong. You can only be so precise when you’re looking through mom, through baby, into a heart. What we heard from the beginning, though, was, ‘Know we can be wrong.’ This is the one case I do not mind doctors being wrong at all. So that was the basis of hope right there” [58]. When faced with the reality of parenting a child with long term complex health needs, parents still held onto their optimism: “That’s when I was like, ‘We can do this. We’re going to do this. We’re going to bring him home. He’s going to do anything he wants in life, and he’s going to outlive us.’ And I’m going to make everything in my body, anything in my will power to make that happen” [49].

Positive thinking varied at different stages of parenting, and the initial shock of the CHD diagnosis led some parents to lose that hope. Despite awareness of children who had gone through the process successfully, some parents struggled to accept the possibility of a negative outcome in case it did not apply to them: “It is hard sometime to stay positive when the specialist is not positive. I had to tell my mom- I cannot hear these success stories anymore because it might not happen for us. So on some days it’s helpful
for me, but on others... it's back and forth” [46]. However, once the child had made it through the surgical procedures, parents reported embracing the positivity in the unknown: “So you learn to take one day at a time with her and enjoy all the little quirks that she does” [57].

**Theme 2: Support systems**

A main form of coping for parents emerged in the theme of support systems; it was present in some form in 19 of the 22 studies. Parents cited various people they called upon at different stages of their journey to seek support, whether this was emotional, practical, reassuring or information providing. While most parents accessed some form of support, they still acknowledged that other people did not truly understand what they were experiencing unless they had gone through a similar experience themselves: “Seek out other parents who have gone through it, and talk with them. Nobody that hasn’t gone through it will understand what you are going through” [51].

**Subtheme 1: Support within the couple**

Parents reported turning to someone close to them for emotional and practical support. Most of the participants in the studies were married or cohabiting, so often shared their experience with their partner who was able to appreciate first hand their distress and provide support. Support from the partner was present in seven of the studies: “The one person who could understand was my husband. He is my best friend, plus the father of my child” [54]. “So, my husband is my greatest spiritual support. Yes...spiritual support. If there weren’t him, I couldn’t have got through it” [61]. Some parents recognised the importance of a shared attitude towards their child, and that it could be difficult to
support each other if this was absent, “If you both agree that you want to keep the baby
and you know, if you have the same goals in general, I don’t think it’s that hard on the
relationship” [57].

Subtheme 2: Support from peers and family
For many parents, close family, particularly their child’s grandparents, became an
invaluable source of support to help parents cope. This consisted of emotional support:
“My and my father have never been close before, but now he gives me all this support,
he told me that he is proud of me. For him to actually say this made me feel really
better” [46]. But it also consisted of practical support to parents whenever they needed
it: “...even just going to the grocery store, or washing my little boy, or not asking me what
I need, but just looking around the house to see we need laundry to be done. That’s one
thing that my mom is good at- she does what needs to be done without asking a hundred
times. I don’t want to tell you, just notice and do it!” [46].

Many parents made connections with others in similar situations through support
forums, and described how they gradually became friends through a shared
understanding: “We joined the Association for Children’s Hearts to look for like-minded
people . . . it’s not so often you run into a family with children with heart defects . . . but
through this association we gained a great deal. It felt like it was, yes, the ability to be
able to keep the head clear in some way” [52]. “The ‘what ifs’ were the big thing. But
once I talked to [friend] and learned and listened to her calmness and her organization of
things—how to prepare, how to function at home —just listening to what she had to go
through, prepared me. So, just asking all the questions and the “what ifs” just calmed me
and helped me deal with it” [59]. For some parents, the anonymity and availability of
online support groups helped them to cope in a way that suited them: “I do not utilize in-person groups… however, I love my online family!” [51].

**Subtheme 3: Support from professionals**

This subtheme was present in eight of the studies, but also refuted in four. Professionals held the position of expert at the start of parents’ journeys, and so were the first providers of information and advice. Parents reported that the honesty, reassurance and information that professionals provided helped them to understand their child’s condition more, and in turn cope better with what they faced as a family: “Every day we started to cope a bit better because we got enough information from the doctor” [55]. “We were very upset initially till we saw the cardiologist. Everyone there was positive about the whole thing. Because of them, we had a great pregnancy” [44].

However, some parents did not feel that professionals helped them to cope, because they felt unsupported by medical staff: “We didn’t have the time or the knowledge to ask everything we wanted to. We were still kind of blown away and it was just information overload on top of it . . . We got a hand drawn heart on a napkin with everything that was wrong . . . it’s like, “Really? You couldn’t have gotten a model of the heart to explain everything to us?” [47]. Some professionals seemed to strike the wrong chord with parents, offering unsolicited sympathy: “I found myself, even within just a few weeks [of the diagnosis], getting mad at people….A genetic counsellor wanted to give me a big hug and [said], ‘I’m so sorry.’ And I’m already kind of getting defensive and getting mad because I’m like, ‘She’s still a baby, and we’re still happy to have her.’ It made me feel like I’m already a parent of a kid with special needs. And I’m not apologizing for my kids, so don’t pity me, don’t pity my kid” [49].
**Subtheme 4: Support through faith and spirituality**

This sub-ordinate theme featured in almost half of the studies. Parents turned to faith, religion and often prayer to call upon a ‘higher power’ for support, and felt comforted when procedures were successful, attributing this to divine intervention: “What kept me calm in the pre-operating area was praying” [44]. “I have faith in god and that’s the only way my husband and I can go through this. My family and I spent a lot of time praying and that helped calm us down” [46]. Parents managed uncertainty around the birth outcome by turning to faith as well, putting their faith in their doctors: “We’ll just let God, put it in His hands and let the good doctors take care of it. That’s about all we can do” [59].

**Theme 3: Parental management**

This theme includes strategies and styles that parents adopted and wove into their everyday lives in order to adapt to and minimise the impact of CHD on family life.

**Subtheme 1: Striving for normality**

Nearly all of the studies in this review referred to a sense of gaining or maintaining normality, and not turning the child’s condition into a reason to treat them differently. Parents who held this attitude reported coping with the diagnosis by not acknowledging it in their day-to-day life: “Raise the child as if they are going to live. Do not treat them as if they are going to die. ... Do not disable them by using their health as an excuse” [51]. “Actually we usually forget the fact that he had heart disease and that he needed an operation. We considered him normal” [61]. This attitude was reinforced by professional advice to new parents: “They never told us, ‘Take this baby home, coddle her, and never
let her do anything.’ That was not how her doctor thought this worked. Let her do what she wants to, let her live a life. If it’s two months that she lives, fine; if it’s two years that she lives, fine. Let her live that life and that’s the way we felt, too” [64]. When offering advice based on their own experiences, the same ideas were given for others to consider: “Live normally. Let your family continue on as normal as possible, because the rest of the world is not going to give one hoot that this kid’s got this heart defect. So don’t let him use it as an excuse. Think positive; hold the vision of good results, and just, you know, deal with it and weave it into your life and your family as normally as possible” [64].

**Subtheme 2: Becoming experts**

This theme features parents’ reports of preparing themselves and their families for welcoming a child with CHD into their lives, by researching information, preparing their home and getting themselves ready to take on the increased care load of a child with often complex health issues. Parents acknowledged the pressure on them once they left the hospital and had to meet their child’s care needs at home, and how they gradually learned about procedures and checks, becoming experts in their child’s condition: “It was a little scary at first. I checked him numerous times a day. I felt very comfortable ‘cause I had all the equipment I needed at home and the more he stayed at home, the more we got to know him... Being home is different ‘cause you don’t have a nurse that you can just run to... I kind of became his nurse while I’m his mom and actually got more comfortable [as each] week went by” [57].
Subtheme 3: Hypervigilance

Parents described coping with their new responsibility of ensuring the health of their child by increased monitoring and checking for any signs and symptoms that would suggest deterioration in health or delayed development. Some became regimented with the medications so they knew that their child was taking exactly what they needed: “I was a fanatic about the medications, you know, that had to be at – the drops at this time and not a minute after” [48].

Hypervigilance was heightened when parents took their children home and they did not have hospital staff to turn to for reassurance: “You just feel like you have a job. I mean your job is very important, even as just a parent of a normal healthy child but it’s not as stressful. You don’t have to worry about did they get their medicine? Are they going to be okay today? Are they going to stop breathing and turn blue? . . . I’ve got to worry about this NG [nasogastric] tube. Is he going to throw it up today? Is he going to pull it out today?” [57]. Even when doctors had reassured parents they did not need to do as many checks, they still continued for their own peace of mind: “So every day, we wrote down his weight and his [oxygen] sats, his saturations [between first and second surgery]. Now [after second surgery], we were not told we had to do it every day, but we did it anyway, just for peace of mind for ourselves” [48].

Theme 4: Avoidance

This theme contains coping styles utilised mainly in the early stages of CHD, such as when a diagnosis has been made in pregnancy, when parents are given the decision of proceeding with surgery and/or when their child is actually undergoing the surgical procedures. The theme of avoidance covers strategies of parents accepting that they are
not responsible for everything, denial of the diagnosis, and using distraction techniques to reduce the time and intensity of worry.

Subtheme 1: “It’s out of my hands”

Parents reported a sense of allowing big events, such as life-saving surgery, just to proceed without trying to control it too much, because they felt that they could not influence the outcome themselves. They relinquished this responsibility onto the surgeons and left their child in their hands: “Sure, we had to sign a consent and such, but there really wasn’t a choice. I looked at it like: A) We cannot have the surgery and our son will die or B) We can do the surgery and there’s a chance that he will live” [54].

Parents also acknowledged that they were physically unable to do everything for themselves and their families, so eventually handed over some aspects of their roles to others in order to cope: “I am not only a parent, I am apparently also just a human with limitation, and I cannot go around the clock [sic] and at the same time doing my job... as it has been for me, my mother was babysitting, so I could go to sleep or work” [52].

Akin to the theme of support through faith, some parents reported believing in handing their child’s fate over to a ‘higher power’ and trusting in God’s plan for them: “I don’t know if I believe in destiny or if I just have a lot of faith, but when there’s nothing else you can do, I had to believe that whatever was supposed to happen, would happen” [54]. Whether the outcome for the child was positive or not, parents reported: “For whatever reason (I thank God), on this day <of surgery> I felt like I could take it if our son did not survive. We were doing all that we could to give him a heck of a chance but you can’t fight fate” [54].
Subtheme 2: Privacy

This theme related to parents managing by not sharing information about the diagnosis; for example, limiting how much they told people who asked about the pregnancy: “So there’s nothing to be ashamed of. And we know that. But we also know that not everybody that talks to me and asks how far along needs to know” [49]. This theme continued when the child was born and underwent surgery.

Subtheme 3: Denial/distraction

In the early stages, once parents received the diagnosis of their unborn child’s heart condition, several reported trying to ignore or deny that there was a problem. The pregnancy felt normal to them, but appointments reminded them of the diagnosis. During the long stays in the intensive care unit, parents reported that they needed something to distract them in order to reduce or avoid the worrying thoughts about what could happen: “I've been playing cards here just to get my mind off. I'll go down and sit with my son for a while and then when I cannot sit anymore I'll say to another mom- let's go and play some cards, I feel stressed” [46]. These passive coping mechanisms seemed to help distract parents from the severity of the situation and gave them a chance to recuperate: “As hard as it's been, there were two times that my husband and I have gone, left and ate dinner somewhere. Not fancy, just not in the cafeteria. We even had a beer one night. It felt really good. The next day I was ready to restart dealing with this” [46].
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Discussion

This is the first review in this area which explored parents’ reports of coping with a child diagnosed with CHD; 22 studies were systematically reviewed and their methodological quality was assessed. Four main themes emerged from the synthesis, detailing parent coping. By drawing together experiences during pregnancy, through surgeries and parenting later in childhood, this synthesis provides a more comprehensive understanding of the various facets of coping that vary over time, reported qualitatively using parents’ own words.

Parents who received a diagnosis during pregnancy had the additional stressor and responsibility of making a decision to proceed with the pregnancy knowing that their child would have a chronic condition. This period of time was exemplified by using emotional coping strategies, such as optimism, attachment (or detachment) to their unborn child, and expressing their grief at the diagnosis. Some parents turned to their partners for support and did not tend to invite others to share in this experience because this would expose the reality of their circumstances. Some parents began gathering information to learn about the condition while others employed avoidance strategies so they only had to deal with the condition once they knew the severity [71]. Parents who received a diagnosis at birth bypassed the period of uncertainty during pregnancy, but had to face the reality of having a child with a heart condition requiring surgery when they had been expecting a healthy baby. The period between birth and surgery may have been short, and so parents had to adapt much quicker than those who already had the diagnosis before birth. Unequivocally, parents reported coping with the management of CHD by doing their best to integrate it into their lives without it taking over, and leading normal lives whenever possible. Nevertheless, many parents reported developing
hypervigilance to any symptoms suggestive of deterioration in the health of their child – an adaptive and entirely appropriate strategy. Parents became experts in their child’s condition, learning about their healthcare needs and developing an understanding of what symptoms indicated the need for medical attention and what was just normal for their child.

Findings from this review concur with a review of 29 studies to determine supportive care needs of parents with children with rare diseases [72] in relation to social, informational and emotional needs. Similar findings relating to parental vigilance and information seeking, as well as support from religious beliefs emerged in reviews of other chronic childhood conditions [73,74]. Mothers of children with other foetal abnormalities use similar coping styles to those reported in the synthesis [75,76], particularly the emergence of hope in the face of unfavourable odds [77]. The idea of coping with the condition by treating the child as normal, or maintaining a normal family life, was present in most of the studies, and echoes findings from other reviews of children with chronic illness [78]. However, it is evident that alongside striving for normality, parents remain vigilant and watchful for signs of potential ill health, when their child has a chronic condition [79] which can be appropriate and adaptive.

The findings are also concordant with those of other studies and reviews [20,24] on this research topic in terms of drawing on social support as a strategy for coping. The theme of support was evident although it ranged from support within the couple to the wider family, to seeking out and accessing support from other parents who were going through similar experiences. This provides parents with a shared social identity and enables them to learn from the experiences of others [80], and use of peer support groups has been found to benefit parents across a range of chronic health conditions.
[74,81,82], both online and in person [83,84]. In the quantitative literature, perceived social support has been found to predict family coping [5] and family cohesiveness [24]. A major emergent theme was the use of faith and spirituality to cope with unknown or difficult experiences. Spirituality can support or hinder children’s coping with chronic illness [85], and it has been shown to support parent coping [86].

There is little qualitative evidence in other conditions that supports the use of denial or distraction as a coping style: this is often labelled as dysfunctional or maladaptive on measures [87]. However, in the unique environment of theatre and paediatric intensive care unit (PICU), there is little else that parents are able to draw on.

**Limitations**

As the metasynthesis approach inherently involves a degree of subjectivity, the studies included in this review were examined independently by the research team. To ensure the rigour of the review, a proportion was independently rated to reach agreement around inclusion and exclusion.

While the search strategy\(^2\) was inclusive, using broad terms, MeSH terms and exploded searches if possible, it is likely that by using “qualitative” as part of the search terms, studies that did not explicitly state the methodology using that term were not identified. However, hand searching of reference lists and using Google Scholar enabled the researchers to find additional relevant studies that were not initially picked up in the database search.

The choice of search terms may have potentially biased the search to positive rather than negative coping styles. The definition of coping used to screen papers may

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\(^2\) For more details on search strategy, see critical evaluation paper.
not have identified such styles that could be subjectively considered maladaptive. Therefore, some papers offering evidence for additional themes could have been omitted.

By excluding non-peer reviewed papers, grey literature featuring alternative perspectives may have been omitted. Whilst there was another review of 25 studies by Jackson, Frydenberg, Liang, Higgins and Murphy [24] in this area, there was only an overlap of four papers with our review because the current metasynthesis focussed on the qualitative studies exclusively.

All of the studies included in this review were considered high to medium methodological quality, because those that scored low were excluded. The studies in this review used the qualitative reports of 704 parents from seven countries across North America, Europe, Asia and Australasia, with common findings featuring across studies. Samples were usually drawn from single heart centres linked to the researchers; thus, there might be differences within that country itself based on the provision of care, especially in countries where health care is privatised. Most parents were married or living with a partner, so these findings do not account for single parent families who may face additional or different challenges to cope with. Although this review aimed to explore the parent experience, mothers outnumbered fathers in the studies by 3:1, a common limitation in paediatric psychology research [88]. It was apparent from the demographic data that though studies may aim to explore the “parent” experience, the number of mothers drastically outnumbers the number of fathers in inclusive studies, particularly in one study [51] which excluded the data from fathers because there were so few, resulting in the findings only applying to mothers.
All of the studies used convenience or purposive sampling which is highly vulnerable to selection bias because it may not be representative of the population [89]. Three of the studies [48,56,57] used transcripts and notes from previous studies by the same authors to answer a different research question, removing the possibility to probe and explore prominent issues deeper. Similarly, the studies which used journal entries [54] and surveys [51] lacked this depth of data: as they did not have contact with the parents directly they could not delve deeper into their narrative. However, this approach enabled the researchers to collect data from a larger sample in comparison to using interviews only.

**Clinical implications and future research**

The findings of this review have clear implications for services that come into contact with parents of children with CHD. The results suggest that parent coping begins soon after receiving their child’s diagnosis, whether antenatally or at birth. There is a grief and stress reaction at the time of diagnosis [90] when parents must adjust to the presence of the condition [91]; this varies as parents begin to passively or proactively cope with the condition, and continues throughout on-going health care, developmental transition stages and any hospitalisations and bouts of severe illness [92] which health care professionals need to be aware of in their interactions with families. While professionals may need to use appointments to explain the intricacies of the condition and offer information, parents can find professionals unhelpful if their responses are interpreted as unsympathetic [93]. It may be beneficial to make distractions available to parents during the surgical and recovery period, and give them “permission” to take a break, as they may feel obliged to stay in the hospital even though there is nothing they can do.
Many parents report drawing on the psychosocial support available to them in the form of wider family and friends. While some parents may withhold details of the diagnosis from people outside the family, they could be informed by their clinician of any local or online support groups available should they wish for contact with other families in similar circumstances. Indeed, the literature suggests that parents can cope with the demands of a child with CHD, particularly when they already have children or have learnt how to manage the health care needs of a child with CHD [11] and display a range of styles of adjustment [12]. However, when families exceed their psychosocial resources due to stressful events, whether related to the condition or other life stressors, they may need psychologically minded professionals to notice their need and offer support. It is therefore imperative, when working with children with CHD, to consider the whole family system [94] because their wellbeing and vulnerabilities can have a direct impact on the wellbeing of the child [95]. There is evidence supporting the use of parenting interventions for other chronic childhood diagnoses [96]; group interventions have been developed to specifically support parents of children with CHD [97] which are usually run by a clinical psychologist who could provide additional input for families where appropriate and indicated. These have the benefit of widening parents’ peer support network, hearing stories of survival and strategies for dealing with stress, as well as normalising their and their child’s experiences.

**Conclusions**

Parents of children with CHD are more vulnerable to psychological and social distress, and the strain of the condition can impact the family at different points along the journey from diagnosis into childhood. This review demonstrates that while there are common
themes of coping in parents, individuals employ their own styles and strategies based on prior experience, availability of social support, personal characteristics and beliefs.

Parents who receive a diagnosis antenatally must psychologically adjust having a child with a heart condition when the child is born and make the decision of whether to proceed with the pregnancy. In contrast, parents who only discover their child has a condition at birth face the sudden shock of the healthy baby they expected actually being very unwell and requiring major surgery a few days after birth so the time they have to adapt to their new situation is drastically limited. Their coping styles draw on their existing resilience and support networks. Once children enter the surgical phases, irrespective of the timing of diagnosis, parents share a similar journey ahead, aided by drawing on existing resources within the individuals and their support network. Parents try to maintain a sense of normality, integrating CHD into their lives without it having a major impact except at times of transition and hospitalisation when they must call upon additional strategies or supports to manage this stress.

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Children with Single Ventricle Congenital Heart Defects:

Considering the Parent Experience

Paper prepared for submission to Congenital Heart Disease

(See Appendix 6 for journal guidelines. Please note this journal has no word limit for manuscripts)

Word count: 6460 (main text)

8209 (complete text including tables, figures and references)
Abstract

Setting: Congenital heart defects (CHD) can be detected during routine antenatal scans, but can also go undetected or undiagnosed until birth. Compared to parents with a typically developing child, parents of children with CHD have a markedly different experience, as they navigate the journey from diagnosis, surgery and after-care to the child developing their own independence.

Objective: This study aimed to explore the in-depth experience of parents of children with CHD who had completed their Fontan surgical procedure.

Design: Interviews were conducted with 12 parents who were recruited from a children’s hospital and were diagnosed either antenatally or postnatally with CHD. Interpretative Phenomenological Analysis (IPA) was used to identify themes across parents’ accounts.

Results: Findings suggest that parents experience a period of emotional distress at the point of diagnosis, then find their own way of accepting and integrating the experience of CHD into family life. Through becoming experts in and taking responsibility for the management of their child’s physical health care, they are able to develop their own sense of normality. Challenges arise when their children reach transitional stages, such as puberty or approaching adulthood: parents are often unprepared for the emotional demands that arise at these points.

Conclusions: Accepting CHD and taking control of its management enabled parents to integrate it into their lives without it restricting their activities. Parents regain a sense of control and normality within the family. In this first study of its kind, clinical implications are discussed, highlighting how health care professionals can promote emotional wellbeing and provide support to families with a diagnosis of CHD at significant time points during the patient journey.

Keywords: CHD, Family, Interpretative Phenomenological Analysis, Qualitative, Interview.
Introduction

The incidence of children born with congenital heart defects (CHD) worldwide is 1%\(^1\). There are multiple variations, and exact diagnoses and prognoses differ\(^2\), so parents must adapt to and manage new challenges, from pre-natal to birth, then childhood and adolescence.

Reviews exploring the parent experience in the presence of other chronic childhood conditions\(^3\) report that significant stages, including diagnosis\(^4\), surgery\(^5\), hospitalisation\(^6\) and developmental milestones\(^7\), signified periods of high stress for families, as outlined in the Typological Model of Family Systems and Chronic Illness\(^8\). Transition into adult services required parents to further adjust their role as they supported their child’s independence\(^9,10\). As well as these explicit stages, it is vital to recognise parents’ expertise in and contribution to everyday care\(^11,12,13\), while living with the uncertainty of the illness\(^14\).

Children born with CHD must undergo multiple, critical, life-saving surgical operations (the Fontan procedure) usually within days of birth, then in their first year, and finally completion around four to seven years old, depending on their development and health. Each of these procedures carries risk and takes several hours, leaving parents powerless and facing the possibility of losing their child. Many children face procedures such as catheters in addition to major surgery to correct defects, and must adhere to a medication regimen. The parent experience during surgery and the management of CHD vastly differs from that of parents with healthy children. Parents are expected to understand a condition they had not previously considered, make significant decisions about their future and that of their child, cope with the uncertainty and distress of surgery and become health care providers as well as mothers and fathers. Therefore,
psychological and practical implications must be considered, particularly when there is a poor or uncertain prognosis.

Support is often available in the early years when children are vulnerable to the physical challenges associated with a CHD; however, parents are expected to take on a caring role to support their child throughout childhood. Wernovsky et al.\textsuperscript{15} outline some of the physical limitations, medication regime, aftercare and family burden that remain once the Fontan procedure has been completed.

Recent studies\textsuperscript{16-18} emphasise the higher incidence of psychiatric comorbidity in adolescents with single ventricle CHD, and the importance of providing psychological and psychiatric support once the physical needs are managed. Studies\textsuperscript{19-26} have demonstrated that parents experience higher levels of stress and mental health difficulties than those with typically developed children. However, parents have reported coping styles to manage this stress and these strategies can vary over time from diagnosis to surgery and beyond. In a review of 22 qualitative studies\textsuperscript{27} parents reported coping with CHD by integrating it into their lives, and utilising additional strategies or sources of support at times of heightened distress.

A 2015 mixed method systematic review\textsuperscript{28} reported the impact of CHD on parents’ psychological health, family life, parenting challenges and family-focused interventions. It highlighted the need for qualitative research into understanding the parent experience over time. This lack of research was partially addressed in terms of the emotional experience from diagnosis to surgery\textsuperscript{29,30} and early years\textsuperscript{5,31}, but of course the parent experience continues beyond this time. Fathers’ experiences have been explored using Interpretative Phenomenological Analysis (IPA)\textsuperscript{32}; however, no study to date has explored the experience of individual parents, whether mothers or fathers, qualitatively. The aim
of this study was to explore the lived experiences of parents who have children with a diagnosis of single ventricle CHD, from diagnosis through to their childhood and adolescence.

**Methods**

Ethical approval and research governance was obtained from the local NHS Research Ethics Committee (REC) and Health Research Authority (HRA) for this study (reference 17/NW/0165, see Appendix 7).

This study employed the qualitative methodology of IPA, because it was consistent with the purpose of the research to gain insight and understanding into the real lived experiences of parents of children with single ventricle CHD. IPA is particularly relevant within health psychology research because it moves away from the biomedical model towards developing an understanding of individuals’ perceptions of their experiences and the meanings they assign to them. It has been used in similar research to make sense of a person’s lived experience of a phenomenon.

**Participants and Procedure**

A convenience sample of 12 participants was used. The cardiac psychology and nurse specialist teams at a large hospital in the North West of England identified mothers and fathers of children known to the paediatric cardiology department. Their children had completed their Fontan surgery at least six months prior to the start of recruitment. Parents had to comprehend and speak English. Families who had left the area or service, and those who were identified by the cardiology team as having an unstable social situation were excluded.
The clinical team sent recruitment packs (Appendices 8-10) to all eligible families on their database. They had no knowledge of who agreed or declined to participate. The lead researcher contacted each parent who returned their consent to contact form (Appendix 10) by telephone and email to arrange an interview at a location of their choice. Study information was reiterated and participants had the opportunity to ask any questions regarding their involvement.

Participants provided written consent (Appendix 11) and demographic information for context (Appendix 12). Interviews were then conducted by the main researcher and recorded using a dictation device. Following consultation with the research team and a review of the literature, a semi-structured interview schedule was developed. It was organised into a sequence of events to explore parent experiences from the point of diagnosis; it covered the process of multiple surgeries, caring for a child with a cardiac condition, family relationships, impact on life and experience of support throughout their child’s life (Appendix 13). The researcher attempted to elicit additional information using prompts for parents to expand and reflect upon the emerging important experiences. Following the interview, participants were debriefed (Appendix 14) and provided with contact details of the research team and relevant support services. A distress protocol (Appendix 15) was followed throughout recruitment and interview.

The Analytic Process

All interview recordings were transcribed verbatim by the lead researcher. Participants were assigned numbers to maintain confidentiality; all transcripts were anonymised. As the process of IPA is iterative, each transcript was read and reread to achieve immersion in the data prior to initial noting. At this stage, semantic content and language was
examined to elicit key meanings and areas of importance for participants. Descriptive and conceptual comments were made to facilitate comparisons between members of the research team (e.g., see Appendix 16). Emergent themes from each interview were noted then analysed together to develop superordinate themes (e.g., see Appendix 17). One interview was discussed in detail by two researchers (ML, DS) who then independently analysed the transcripts and discussed and agreed themes with the third researcher (AW).

**Reflexivity**

A reflective journal was kept from the point of interview through to analysis to record any reflections, comments and subjective assumptions to keep an audit trail and maintain transparency.

It is important to consider the potential effects on reflexivity of the researcher’s social position, personal experiences and political and professional beliefs in order to understand how these can impact on the quality of the research. The lead researcher (ML) is a white, middle class woman with several years’ experience working psychologically with children and their families. This research was conducted as part of her clinical psychology doctoral thesis. DMS is a researcher in health psychology with two children. AW, a mother of two children, is a researcher in clinical psychology, perinatal mental health and parenting.
Results

Participants

Of the 125 eligible families invited, 17 returned consent slips and 10 responded to contact (see Figure 1). Demographics were collected to provide context to the interview and overall sample (see Tables 1 and 2 for details of parent age, sex, ethnicity, marital status and employment status, child’s age, sex, ethnicity, diagnostic information and position in family). Participants ranged in age from 38 to 64 years (mean=49, SD=7.9) and their children ranged in age from 7 to 19 years (mean = 13.2, SD=4.5). Two couples requested to be interviewed as a dyad; they were assigned one participant number (1 and 7) while one couple opted to be interviewed individually and their data was treated entirely separately (8 and 9).
**Table 1. Demographic data for participants**

<table>
<thead>
<tr>
<th>Participant</th>
<th>Age</th>
<th>Sex</th>
<th>Ethnicity</th>
<th>Marital status</th>
<th>Employment status*</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>59/64</td>
<td>F/M</td>
<td>African/Asian</td>
<td>Married</td>
<td>Retired</td>
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<tr>
<td>2</td>
<td>44</td>
<td>F</td>
<td>White British</td>
<td>Married</td>
<td>Employed FT</td>
</tr>
<tr>
<td>3</td>
<td>46</td>
<td>F</td>
<td>African</td>
<td>Married</td>
<td>Employed PT</td>
</tr>
<tr>
<td>4</td>
<td>46</td>
<td>F</td>
<td>White British</td>
<td>Married</td>
<td>Employed PT</td>
</tr>
<tr>
<td>5</td>
<td>52</td>
<td>F</td>
<td>White British</td>
<td>Divorced</td>
<td>Employed FT</td>
</tr>
<tr>
<td>6</td>
<td>57</td>
<td>F</td>
<td>White British</td>
<td>Widowed</td>
<td>Self Employed</td>
</tr>
<tr>
<td>7</td>
<td>50/50</td>
<td>F/M</td>
<td>White British</td>
<td>Married</td>
<td>Employed FT</td>
</tr>
<tr>
<td>8</td>
<td>42</td>
<td>M</td>
<td>White Irish</td>
<td>Married</td>
<td>Employed FT</td>
</tr>
<tr>
<td>9</td>
<td>40</td>
<td>F</td>
<td>White British</td>
<td>Married</td>
<td>Unemployed</td>
</tr>
<tr>
<td>10</td>
<td>38</td>
<td>F</td>
<td>White British</td>
<td>Married</td>
<td>Unemployed</td>
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</tbody>
</table>

*FT and PT denote full time or part time employment*
Table 2. Demographic data for children*

<table>
<thead>
<tr>
<th></th>
<th>Age</th>
<th>Sex</th>
<th>Stage of diagnosis</th>
<th>Diagnosis</th>
<th>Position in family</th>
</tr>
</thead>
<tbody>
<tr>
<td>C1</td>
<td>17</td>
<td>M</td>
<td>Pregnancy</td>
<td>Tetralogy of fallots and dextracardia</td>
<td>3rd of 3</td>
</tr>
<tr>
<td>C2</td>
<td>7</td>
<td>F</td>
<td>Post birth</td>
<td>Double inlet left ventricle transposition, hypoplasia, subaortic stenosis</td>
<td>2nd of 2</td>
</tr>
<tr>
<td>C3</td>
<td>19</td>
<td>M</td>
<td>Post birth</td>
<td>Hypoplastic left ventricle, ventricular septal defect, mitral atresia, pulmonary atresia</td>
<td>1st of 2</td>
</tr>
<tr>
<td>C4</td>
<td>12</td>
<td>M</td>
<td>Post birth</td>
<td>Hypoplastic right heart syndrome with pulmonary atresia</td>
<td>1st of 2</td>
</tr>
<tr>
<td>C5</td>
<td>16</td>
<td>M</td>
<td>Pregnancy</td>
<td>Hypoplastic right ventricle, pulmonary atresia, tricuspid atresia and ASD</td>
<td>Only child</td>
</tr>
<tr>
<td>C6</td>
<td>13</td>
<td>M</td>
<td>Post birth</td>
<td>Hypoplastic left ventricle, pulmonary atresia, total anomalous pulmonary drainage, atrial septum defect, ventricle septum defect and organ transposition</td>
<td>Only child</td>
</tr>
<tr>
<td>C7</td>
<td>16</td>
<td>M</td>
<td>Pregnancy</td>
<td>Atrial septum defect, ventricle septum defect, subaortic stenosis, transposition of great arteries, double outlet right ventricle</td>
<td>3rd of 4</td>
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<tr>
<td>C8</td>
<td>7</td>
<td>F</td>
<td>Pregnancy</td>
<td>Tricuspid atresia</td>
<td>2nd of 3</td>
</tr>
<tr>
<td>C9</td>
<td>7</td>
<td>F</td>
<td>Pregnancy</td>
<td>Tricuspid atresia</td>
<td>2nd of 3</td>
</tr>
<tr>
<td>C10</td>
<td>12</td>
<td>M</td>
<td>Post birth</td>
<td>Hypoplastic left heart syndrome</td>
<td>1st of 4</td>
</tr>
</tbody>
</table>

*Ethnicity data were collected but removed to ensure participant anonymity
Figure 1. Flow diagram outlining recruitment process

Potential participants identified as having had Fontan procedure between 2004-2016 (n = 180)

Potential participants screened by Cardiac Nurse Specialists/Consultant Clinical Psychologist/Assistant Psychologist (n = 180)

Potential participants deemed eligible to approach as per inclusion/exclusion criteria (n = 125)

Care transferred to different hospital (n = 28)
Transitioned to adult services (n = 15)
Deceased (n = 8)
Lack of English speaking/understanding ability (n = 2)
Social situation (n = 1)
Emigrated (n = 1)

Research packs posted out (n = 125)

Return of postal reply slip (n = 17)

Email/telephone contact made (n = 10)

Did not respond to telephone or email contact (n = 7)
Interviews

All interviews took place between August and September 2017; nine at the participants’ homes and one at their workplace. Each interview followed the same schedule; however, the interviews varied depending on the resulting narratives. Interviews lasted between 34 and 135 minutes, with a mean interview length of 79 minutes (SD= 29.5).

Themes

Transcripts were analysed (see Appendices 16 and 17 for an excerpt) and three super-ordinate themes were identified, with further subthemes offering more nuanced detail, illustrated in Figure 2 below. The super-ordinate theme of ‘Super Parents’ contained four subthemes of parental responsibility, hypervigilance, becoming experts and managing emotional impact. The super-ordinate theme of ‘Striving for Normality’ contained five subthemes of integrating the condition into lives, treating child as siblings/peers, celebrating achievements, recognising limitations and practical challenges to normality. The super-ordinate theme of ‘Accepting CHD and their Role’ contained six subthemes of timing of diagnosis, management of CHD, the ‘hidden condition’, ‘wouldn’t change a thing’, need for support, and letting go of responsibility.
Figure 2: Diagram of Themes

Super Parents
- Becoming experts
- Hypervigilance
- Managing emotional impact
- Parental responsibility

The Parent Experience
- Striving for Normality
  - Integrating CHD into lives
  - Treating child as siblings/peers
  - Celebrating achievements
  - Recognising limitations
  - Practical challenges to normality

Accepting CHD and their Role
- Management of CHD
  - Hidden condition
  - 'wouldn't change a thing'
  - Timing of diagnosis
- Letting go of responsibility
- Need for support
**Super-ordinate Theme 1: Super Parents**

Parents reported a sense of being in control of the important decision making process regarding their child and the management of an unfamiliar condition. Upon taking their child home and commencing parenting, they were responsible for the care of their child outside the protected ‘bubble’ of the hospital environment. Despite a level of uncertainty, parents ‘did what they had to do’ for their child.

*Becoming experts*

Parents went through a process of learning about their child’s specific diagnosis, its symptoms and management in order to provide health care as well as parenting their child. Much of the information was provided at the hospital at the point of diagnosis was difficult to retain, because it was an emotional time, particularly for first time parents.

*I felt very young, I wasn’t young, I was 30 odd, but I felt very young
and by the time we left [hospital], I felt like a grown up! (Parent 4)*

*We know nothing. I don’t know how to deal with a cardiac child. It’s bad enough a kid, keeping it alive, I’ve got a cardiac that’s even, double the trouble! (Parent 10)*

However, over time parents learned what was typical for their child and began to master the medical needs of their child, reducing the need for hospital visits and increasing self-confidence in their knowledge.
You will quickly become the expert on your child’s diagnosis. You will know more than any doctor that ever treats him and you will be right. (Parent 6)

Parental responsibility

Half of the parents interviewed learned of their child’s condition during the pregnancy and faced the responsibility of deciding whether to proceed and opt for the Fontan procedure, palliative care or termination. Parents who decided to proceed with pregnancy, knowing the associated health risks, reported feeling directly responsible for their child’s difficulties. Regardless of when parents learned of the diagnosis, they still questioned whether they were responsible in some way as a result of their actions, which caused feelings of guilt and distress.

I mean the feeling was guilt. I mean I was overcome by I suppose a really profound sense of guilt about having inflicted this on a child.

(Parent 8)

I was annoyed, cos I didn’t smoke, I didn’t drink. I didn’t do anything, I didn’t eat the foods that I wasn’t supposed to eat, you know... I just don’t understand why I got it. There was nothing I did to say, oh, it could be that... It could be that... There’s nothing, absolutely nothing.

And it’s just like, just one of those things... (Parent 10)

On leaving the hospital, parents took on the responsibility of ensuring their child received the best care and treatment possible, so became advocates who fought
for their child’s wellbeing. This level of responsibility often meant changing working patterns and social activities to meet care needs; they were reluctant to leave their child with others due to the burden it may present and the anxiety they would feel by not being in control.

We still struggled with handing him over in that when he started playgroup and we left him, we used to sit in the car outside. And we did for 12 months! (Parent 4)

**Hypervigilance**

A common recurring narrative amongst parents was the notion of ‘wrapping their child in cotton wool’, with parents regarding themselves as overprotective against the risks and dangers of the world. This appeared to be more prevalent in the early years, particularly in between surgical procedures when children were susceptible to infection and breathlessness, so parents had to be constantly available, even after surgeries were complete. This need to protect often put restrictions on parents’ social and occupational activities, because they strived to always be watchful, and available whenever they might be needed.

I never left him. He wasn’t left alone. Where normally you can go and put the kettle on, he was getting carried with me, so I could keep an eye on his lips, his fingers. (Parent 10)

So you could never relax because you never knew when you’d be called because he’d you know, thrown up, whatever, something, you know, every week. (Parent 6)
Manage emotional impact

Throughout the parenting experience, parents were faced with new emotional challenges. One of the most significant emotional periods was during the surgical procedures, when they felt powerless, putting their trust in the surgeons, and consenting to surgical procedures despite varying odds of survival. For some, there was a sense of detachment until they knew their child would survive.

I kept thinking... what colour flowers do you have at a baby’s funeral?

(Parent 10)

If he’s gonna die, I want it to happen now, because I didn’t know him.

(Parent 5)

Parents reported that these thoughts were very difficult to experience; they did not want to share them with others for fear of how they would be perceived. In hindsight, they reported accepting the thoughts and feelings as necessary to cope with the difficult situation they faced.

Super-ordinate Theme 2: Striving for normality

While acknowledging the life changing nature of having a child with CHD, parents reported trying to lead, and give their child, as normal a life as possible. As they mastered management of the condition as a family, they were able to find their version of “normal” and integrated CHD into their everyday lives, reflecting that despite being a serious condition, CHD was a relatively small aspect of their child’s life.
Treating child same as siblings/peers

Half of the parents interviewed already had children and therefore were able to distinguish between symptoms of CHD and typical child behaviour and needs. Parents did not want their child to be treated differently to his/her peers because they did not see the CHD as a disability. Their concern was based in the present but they also considered future functioning, and how to minimise the impact CHD had as their child grew up in order to minimise their child feeling different, special or excluded.

*He’s a normal child, his problem is inside, it’s not outside, and not visual, so we thought, no, why should we send him to a special needs school?* (Parent 1)

*I think you’ve got to be careful with all that, with kids who have got problems of any sort really, otherwise they grow up thinking they’re special, and you know, they need to be given extra attention and... I’d hate him to be like that.* (Parent 5)

Those with other children reported feeling compelled to treat their children alike. Their other children provided a comparison highlighting where their child with CHD struggled in terms of ability or development, so may inevitably have had some special treatment.

*We’re probably fortunate in that, he wasn’t our first, and we didn’t have the time to wrap him up in cotton wool!* (Parent 7)
I’ve tried to keep him as normal, in inverted commas, as possible, you know... I have let him climb a tree. Yeah, [husband]’s followed. I have let him go high on a swing, [husband]’s at the back, I’m at the front, in case he falls off... You know, we have let him climb the rocks in the park, again, [husband]’s with him, two steps behind him.

(Parent 10)

Understanding limitations

Parents were able to recognise areas in which their child inevitably struggled in comparison to their peers; this included physical, social, or emotional limitations and also developmental delays that made them feel different to other parents.

If she gets a cold, it does wipe her out, just takes her a little bit longer to recover than you know, someone with a normal heart. (Parent 2)

Reaching milestones... social media is a nightmare, cos you know, the day [child] said his first word, he was probably three and a half. You know... everyone’s like, oh my child did that ages ago. (Parent 4)

Recognise achievements

Parents were advised that their child may be delayed or not reach the milestones of speech and walking, so they were elated when their child spoke their first words and took their first steps. Seeing their child exceed expectations gave parents reassurance and optimism that CHD was not impacting their lives, which offered hope for the future.
Speech wise we realised, you know, he was doing absolutely fine! You know, 50 words by 18 months, it seemed to be you know, absolutely brilliant. (Parent 6)

When you think yeah, actually she’s got this far, she’s still alive, and she’s running around and stuff. So there were certainly times when you think gosh, that’s wonderful, that’s fantastic.

(Parent 8)

Parents reported significant moments when they recognised their child leading a normal life and a typical childhood, such as starting school and participating in sport, as if there was no underlying heart condition at all. They spoke with pride about how their child had overcome the limitations of CHD and how typical childhood achievements were more significant as a result. Parents whose children who were not as physically able were proud of the positive influence they had on friends and family who saw how CHD impacted their lives.

She got chosen to represent the school. Now it’s only a small school, but to represent her class in a sporting event. And things like that you think, yeah, that’s great, I mean that’s a big thing, and you want to make her realise, without making things too obvious... it’s such a great achievement. (Parent 9)

My friend’s husband has just cycled from Lands End to John O’Groats... for Little Hearts because he’s seen the effect... Just very
touching that [child] can have that influence on other people... It’s just very, it’s nice, it’s a proud moment. And you don’t have those proud moments over winning at sports day... so it’s nice to have them. (Parent 4)

Parents who devoted most of their time to their child’s early years described a sense of gratitude and liberation after the final stage of the Fontan procedure. Their child did not rely upon them as much and gave their family a new lease of life without the restrictions they had previously encountered.

He actually walked to the beach, and back... it was just like we’d been let out of prison. Because we realised he could walk that far, and it opened up the possibility. (Parent 6)

Practical challenges to normality

Whilst striving to maintain normality in their lives, parents reported other issues that made life more difficult for the family, such as caring responsibilities, relationship problems, long hospital stays, mental health difficulties, being unable to work and managing finances to support their family. Such matters could be stressful for any family, but for these parents they added another layer of complexity to an already challenging situation.

In the space of a year, we got married, had a cardiac child and he had an operation, so it was like a bit of a crazy time, anyway, it was a bit of a whirlwind romance for us, and then buying a house, having a kid,
and everything was just so... It was like a pressure cooker, so having
[child] with his condition as well just, popped the lid. (Parent 10)

He is not technically, legally disabled, he comes under a different
section called critical medical needs, and therefore does not qualify
from assistance from [social services]. (Parent 6)

Super-ordinate Theme 3: Accepting CHD and their Role

Parents all gave similar accounts of shock and surprise at the unexpected CHD
diagnosis. They described the contrast of elation and excitement about their baby,
with the sadness and fear from the realisation that their precious child had
something wrong them. Parents faced an uncertain prognosis, and had to consider
the risks of surgery in their role as decision makers for their child. Parents
described a process of coming to terms with and managing the condition, gradually
accepting it into their lives.

Timing of diagnosis

Half of the parents interviewed had the diagnosis before birth, and half after. Those
who received a diagnosis at the 20-week-scan could proceed with the pregnancy
followed by surgery or palliative care, or had the option to terminate due to
uncertainty around the baby’s health. Parents in the former group spoke of the
shock they experienced.

You rather foolishly, I think a lot of people assume that scan, the
anomaly scan, is where they see what sex the baby is, if you want to
Parents who received a postnatal diagnosis had expected their child to be healthy because nothing had been identified at the scan, so it was difficult to accept the diagnosis, but in retrospect, they were not sure if they would have wanted to know earlier.

*When I was told, it actually broke me down. Because I couldn’t really pinpoint what happened. I don’t know what happened, that was the question up to today no-one can answer. What happened?* (Parent 3)

*With the 20-week-scan, there’s swings and roundabouts with knowing and not knowing. I wish I knew for the whole, like, that wouldn’t have been a massive shock... And then if I knew, I’d have spent 20 weeks worrying, what’s gonna happen to him when he’s born?* (Parent 10)

**Management of CHD**

Parents all spoke stoically of accepting the management of CHD as part of their lives. A recurring topic was how parents share information with their children about their CHD, and answering questions they had. Some opted for an early, honest approach, while others were unsure how to approach the subject due to its inherent uncertainty. Parents recognised that their child had to understand their own condition so they were aware of their own limitations, but also because they...
would be responsible for managing it in the future. This decision conflicted with wanting to protect and treat them normally, causing some parents to struggle to find the best approach.

_Tell him the truth, cos it’s gonna come out, and he’s gonna be like, why have you never told me this? And he’s gonna know, he needs to know, cos he needs to know his own limits, because if he tries to keep up with everyone in the playground, he’s gonna falter, he’s gonna fall._ (Parent 10)

_It’d be much, much easier if it was a prognosis that you were able to say well you’ve had this, this and this done, and you’re fixed and that’s it._ (Parent 7)

**A hidden condition**

Since CHD is not immediately noticeable, other people may not realise or understand the limitations, complexities or prognosis, and they simply see a child who has had some operations when they were younger but are well now. Parents reported feeling frustrated with family members and others who made false assumptions about their child.

_They see her... running around and jumping around on kind of walls and all sorts, they’re like, oh she’s grand isn’t she? She’s absolutely fine... there’ll be no need for anything else. You know, as if the heart has just grown back or something. I said no, she’s still gonna need a_
transplant at some point, you know. It’s kind of this, don’t really understand the gravity of it. (Parent 8)

“Wouldn’t change a thing”

All of the parents reported a sense of stoicism, acknowledging that they would not choose to burden a child with the pain of CHD or to have to go through surgery, but they would not change their child either. Furthermore, they accepted their role in parenting and caring for their child as something they just had to do, and gave it their full effort.

That’s what parents have to do... put their children first and they’ll be alright, you know, they’ll grow up slowly... the disease is gonna be there, it’s not gonna be gone, I mean they can’t do a heart transplant til somebody has a heart and lung on the right instead of the left...

They’ll have good days and bad days, obviously... you just have to cope with that. You know, best way you can. (Parent 1)

I would never get rid of you regardless of your condition, and if I could have my time all over again, I would... if I could have him without his heart condition, of course, for him. But not for my sake, for his sake, so he doesn’t have to have the ops. But [child]’s not [child] without his heart. And that’s all I’ve known. (Parent 10)
Many parents reported a change in perspective having their child with CHD, recognising priorities and changing aspects of their lives things that did not fit with their newfound values.

*Accepting support*

There were mixed reports of the experience of support between the parents. There was a sense of needing an ‘ally’ to cope with the whole experience, whether that was their partner, parents or a close friend. Some parents appreciated contact with other families in similar situations who understood their experience. However, this was not the case for all participants, because some parents preferred to rely on existing support networks and spent their time on typical family activities:

*The support groups, they have their place, and there’s all sorts of activities that you could go on, weekends away and days out... but in all honesty, I don’t want to spend my time while [child] is well with lots of other parents and all their children, with similar conditions, talking about what they’ve got wrong with them.* (Parent 7)

During the early stages, most parents had access to the cardiac nurse specialists who were available by telephone to answer questions and offer reassurance to parents who were learning to care for their child. Provision of practical support that made their lives easier was appreciated, and the local hospital was reported as providing beneficial educational sessions for parents of children approaching secondary school. As the child approached adolescence, other unexpected
challenges arose within families, and parents acknowledged the need for support around psychological issues they had not anticipated.

*I used to go to the laundrette but then after [child] was born and because he was being that sick and all that... so that’s the help we got from Family Fund Trust. He used to sweat a lot, on medication... they gave us a washing machine, paid for the driving lessons which was very, very good.* (Parent 1)

*Getting the parents together and just sort of talking about our experiences and being told what to expect and what school should be doing, just makes you feel much more confident.* (Parent 4)

However, there were several reports of feeling unsupported, often due to their geographical location or lack of services available to the family.

*I think social services should get involved, and see, you know, is that family doing alright? Are they coping financially? Emotionally? Do they need any help? Respite? You know, we can’t go on holiday – I’ve not been on holiday... can’t go anywhere, you know!* (Parent 1)

*Letting go of responsibility*

Some of the parents had children transitioning into adulthood; they reported concerns about letting go of their responsibility over their child and allowing them independence.
It’s worse now, you know my little apron strings are getting frayed.

They haven’t quite been cut but they’re getting frayed! And he’s unravelling them every day! (Parent 10)

I’m desperately trying, I would love for him to become independent and it’s like trying to shove a chick out of the nest, because he’s got so used to the fact that I’m always there, will always support him.

(Parent 6)

Discussion

This was the first study to explore the experiences of parents of children with CHD from pregnancy and throughout childhood to adolescence, and it highlighted three main themes. Parents went through processes of accepting the diagnosis, understanding that it was a relatively rare life-long condition requiring constant management, for which they would need to take control through learning about the physical needs, surgical procedures, regular monitoring and medication. Despite this, parents established their own versions of normality in the face of adversity, developing their own narrative to integrate CHD into their lives, being aware of its restrictions but celebrating when their children overcame expectations. All parents spoke of making sacrifices including employment status, family time and social activities, but wholly accepted these as part of their role as parents to do the best for their child.

Parents reported shock and distress at their child’s diagnosis of CHD, regardless of whether this was at birth or antenatally, suggesting both are similarly
difficult. This event presents an opportunity for psychological support both during pregnancy or hospitalisation due to the multiple elements that parents must prepare for and the ‘rollercoaster’ of emotions they experience. Although there were fewer reports from fathers in this sample, those who participated did report emotional distress and so should be offered support alongside mothers, as this is often neglected. Fathers’ responses in this study differ from those in other studies which emphasised the importance of remaining strong and taking on a caretaking role in the family, hiding emotional distress and not feeling confident to express their fears. Fathers in the current study openly experienced distress and, in some cases, struggled to cope more than their female partners. All parents mentioned the wealth of information they had to learn, and the difficulties in processing what medical staff were telling them. Once their child was born, parents seemed to accept CHD and the responsibilities that came with it as inevitabilities that they had to adapt to. Some spoke of wishing that their child did not have CHD, for the sake of the child who had to endure the pain of surgery, but otherwise would not want to change who they were as a result of the condition. Although parents who received an ante-natal diagnosis had additional time to prepare and educate themselves, there was still a level of uncertainty around the birth, and the risk associated with surgery was applicable to all. As parents became more knowledgeable and skilled in the management of their child’s CHD, however, this can lead to a feeling of isolation. It became apparent that families with healthy children did not have the same demands or understanding of the complexity and risk these parents had to manage. Therefore, contact with services or peers to normalise families’ experiences can be beneficial to reduce these feelings of
isolation should they choose to participate. However, some felt that while these played an important role, they preferred to maintain their version of a normal life by socialising with friends without the emphasis on CHD.

Control was a major recurring theme throughout the interviews. Parents strived to gain control over CHD for the wellbeing of their child so that it did not restrict their lives. Through establishing control, they were able to create their own version of normality. This differed between families, mediated by context, such as other siblings, severity of CHD, support available and individual traits. The reported difficulty in relinquishing control of CHD supports previous research on transition of care from parents to their adult children and highlights the need to support parents to facilitate their child’s transition.

The challenges reported by parents reflect those reported by Coffey including ‘living worried, carrying the burden and taking charge’, which are found in other chronic childhood conditions. These findings demonstrate common experiences in parenting in the presence of a chronic childhood condition, and findings are very similar to the experiences of parents of children with cancer, likely due to the severity and life limiting nature of the condition. The major difference lies in the experience of hospitalisation and planned surgery that children must undergo, which presents risks and uncertainties parents have to endure and these experiences may contribute to higher levels of psychological distress, compared to parents of healthy children and children with other conditions.

The stages described by parents in this study can be understood using the Family Systems Typological Model. This conceptualizes chronic conditions by
onset, course, outcome and degree of incapacitation, which are hypothesised to be significant factors for the family to manage. The model, which categorises CHD as ‘constant, non-fatal, acute, non-incapacitating’, proposes that each phase of an illness has its own psychosocial tasks which require strengths, attitudes or changes from a family. The family needs to create a meaning for the illness while feeling in control, grieve the loss of their expected healthy child, adjust to accept their new roles, work together to cope with crises, and be flexible regarding an uncertain future.

Limitations

Findings are reported from interviews with 12 parents (including two couples) talking about their own experiences, so must be interpreted with caution in relation to transferability. The two couples (P1 and P7) requested to be interviewed together, so their interviews were analysed as one parental “unit”, potentially detracting from any individual differences within the couple. Given that the study used a voluntary convenience sample, there is a risk of sample bias, because it is likely that these participants volunteered to partake in order to tell their story. Those who declined might have had a different experience. All participants had children with single ventricle CHD, but within this diagnosis their conditions differed, as shown in Table 2. Thus, the sample was more representative of the clinical population\textsuperscript{48}, while lacking the homogeneity ideally required for IPA\textsuperscript{38}. However, this study benefitted from a large sample size for an IPA study which provided a robust overview of the parent experience. The resultant themes
demonstrate commonalities across the interviews, providing insight into the parent experience.

**Research Recommendations**

Similar to another study by Gower et al\(^{32}\), this study also highlights the need for more research with fathers of children, not only with CHD but all chronic conditions, because they are under-represented in the literature, as shown in the study characteristics of Paper 1. Within this study, three couples were interviewed but only one opted to be interviewed individually (P8 and P9), because they felt their experiences differed sufficiently to indicate this. As this approach provided insights into different perspectives on the same phenomenon\(^{49}\), future research could explore couple experiences of having a child with CHD to compare findings and identify similarities or differences. By understanding and acknowledging different experiences and approaches in the same family, healthcare professionals are able to offer tailored advice and guidance to both parents.

**Clinical Implications**

Healthcare professionals are obliged to provide sufficient information to families at times of diagnosis\(^{50,51}\); however, it is apparent that parents must come to terms with it before taking charge and managing the responsibility. Parents in this study generally appreciated the honest, sometimes blunt but realistic communication of the prognosis and risks associated with CHD; however, it cannot be assumed, particularly without prior knowledge of the family, how they will opt to proceed with the pregnancy. Parents understand that cardiologists have limited time to spend explaining the intricacies of their child’s condition, but appreciate having a
cardiac nurse specialist reiterate explanations, often in lay terms, and be available to answer queries they may not feel confident to ask. Those parents who may not feel as confident should be supported to seek clarification, record appointments and ask for additional input as they might not be forthright in doing so.

There were multiple stages, as described in Rolland’s model of Typology when parents benefitted from additional support from their care team: on discharge from hospital, when parents began caring for their child at home, they often sought guidance from cardiac nurse specialists to clarify what was “normal” for their child. This was even more important for first time parents who did not have an existing frame of reference for typical infant behaviour, and they reported regular and reliable support from the cardiac team. Therefore, psychologists could play an important role in providing cardiac nurse specialists with education or training to help them notice signs of mental health conditions in parents. Transition points represented potential key periods for additional input from health care services; for example, entering puberty or moving into adult services. When children began presenting with emotional or mental health needs, parents recognised the need for specialist psychological input for advice, assessment or intervention, either directly with their child or indirectly. Parents would benefit from normalisation in order to recognise and validate the difficult emotions they experience around significant events, especially if they do not know other parents with a shared experience. Clinicians should anticipate these potential stages when priorities and needs shift for families, and tailor their level of support or interventions accordingly. A 2015 review proposed that the supportive care needs for parents of children with rare diseases were mainly social, followed by
informational and emotional. Therefore, it may also be advantageous for families to be advised of support groups and forums to provide peer support and reduce feelings of isolation. Pye and Green propose a homecare manual for new parents to educate themselves around the condition during the stressful early stages when parents are managing their child’s medical and physical needs, which could begin to address the need for information. Families could be provided with practical support through referral to services or charities which offer assistance with social and financial support, such as understanding what benefits they are entitled to and assistance completing applications required for these. As recommended by Jackson et al., broader interventions such as education or group sessions could be delivered by a health psychologist, while targeted specialist interventions for parent or child may require a clinical psychologist or family therapist, especially if parents have existing mental health difficulties.

Conclusion

This study explored the parent experience of having a child with single ventricle CHD, from diagnosis, through childhood, until they transitioned to adulthood. It found that accepting the condition and its management enabled parents to integrate CHD into their lives without it restricting their child’s or their own activities and regain a sense of control and normality within the family. Transition points such as starting nursery, final surgery, puberty and reaching adulthood are key time points when parents may struggle with their changing roles and the gradual shift of responsibility and independence onto their children. This has
implications for supporting parents to educate their children in terms of the management and long term prognosis of their condition.

Acknowledgements

The authors would like to thank all the participating parents who openly and kindly shared their experiences of living with a child with CHD and the cardiac team at Alder Hey Children’s Hospital for support with recruitment.
References


Paper 3

Critical reflections on the systematic review and empirical research

Word Count 3324 (main text)
4042 (including references)
Introduction

The preceding papers are related through their exploration of parent coping and the broader parent experience of having a child with CHD. The aim of this paper is to provide a critical and personal reflective account of carrying out two distinct, albeit related, pieces of qualitative research. This paper is split into two parts with critical appraisal and personal reflections interwoven throughout. The first part pertains to the intricacies of conducting a meta-synthesis; the second part contains a critical appraisal of using a phenomenological approach (Interpretive Phenomenological Analysis; IPA) and the interview process. The clinical applications and future areas for research will also be discussed.

International statistics for the prevalence of CHD have been reported in the systematic review and empirical paper; however, in the UK 1 in 180 babies are born each year with some form of CHD (Townsend et al., 2013), with more being diagnosed later in life. NHS England (2016) specifies standards that specialist paediatric cardiac centres must follow to provide best care for children diagnosed with CHD and their families. Approximately 80% survive until adulthood, and research (Kovacs et al., 2009) shows a high prevalence of mental health problems in adolescents with CHD compared to controls (Westhoff-Bleck et al., 2016). In addition, parent mental health and emotional wellbeing is impacted (Kolaitis, Meentken, & Utens, 2017), indicating the need for research to understand why and when families may need additional support. The cardiology team at Alder Hey identified this clinical need and approached the University of Manchester to develop the research project.
Part 1: Systematic Review

Developing the Research Question

A scoping search found several reviews since 2015 relating to parent coping with CHD; however, none of these used a purely qualitative approach. Other reviews exploring coping used samples consisting of parents of children with other chronic conditions. While these studies formed a useful comparison, it was not possible to extract findings specific to CHD. There was a mixed method review published in 2015 (Jackson, Frydenberg, Liang, Higgins, & Murphy, 2015), but they only included four qualitative studies, so the parent voice was not represented. Therefore, by focussing on CHD and qualitative studies only, this review would provide new and useful findings.

Systematic Search

The search strategy, detailed in Figure 2 and Appendix 3, was developed using SPIDER (Cooke, Smith, & Booth, 2012). However, this tool has been criticised for lower sensitivity (Methley, Campbell, Chew-Graham, McNally, & Cheraghi-Sohi, 2014). The use of “qualitative” as a search term, rather than listing all potential qualitative methodologies as terms, was intended to be inclusive but actually limited the results depending on the use of the term in keywords. Due to the above restrictions, hand searching was considered necessary to minimise the potential omission of relevant studies. Inspection of why 12 supplemental papers found from reference lists and Google Scholar were not identified by the search strategy revealed that they included specific diagnostic terms (e.g., hypoplastic left heart...
syndrome) which had not been included in the systematic search. While it was expected that using the term “qualitative” would restrict results, in actuality it was the lack of medical terminology. For a novice researcher, it is important to be aware of the possible diagnoses or other specialist terminology so that they can be included in a search.

Limitations of Studies Reviewed

Qualitative studies, by their nature, tend to use small, heterogeneous samples, methodologies and contexts, and therefore generalisable conclusions cannot be drawn from their findings. The studies in this review use different qualitative methodologies. Estabrooks, Field and Morse (1994) suggest that findings of similar research articles may be aggregated in a review such as this in order to develop generalisations. Given that various methodologies were employed in the studies reviewed, it can be argued that original data can be lost in such a synthesis due to differences between authors’ interpretations (Sandelowski, Docherty, & Emden, 1997). This is a general weakness of qualitative reviews. However, systematic reviews are important in providing breadth, rigour and transparency to the literature review process (Mallett, Hagen-Zanker, Slater, & Duvendack, 2012) and qualitative reviews in particular combine rigorous processes to present the collective meaning of the empirical evidence base (Bearman & Dawson, 2013).

Quality Appraisal

All 27 studies from the systematic search were subject to quality appraisal (Hannes, 2011) by the main author using the Walsh and Downe (2006) checklist resulting in
the exclusion of five studies based on low methodological quality. An independent researcher critically appraised 30% of these 27 papers, resulting in 75% agreement in categorisation (Carroll & Booth, 2015). The use of a percentage value to demonstrate inter-rater reliability could be critiqued because this method of calculation does not take into account the element of chance. An alternative and more robust method for calculating agreement would be to use Cohen’s kappa coefficient (McHugh, 2012) which takes into account the possibility of agreement occurring by chance. Landis & Koch (1977) provide a guide to the strength of agreement according to the value of the kappa statistic.

The aims or purposes of the research were clear in all of the 22 studies that were eventually included in the review, and all studies were well contextualised by existing literature. The majority of studies provided details on participant demographics, making it possible to compare the samples by age, gender, ethnicity, income and education; however, this information was not always incorporated into the discussion, rather it was referred to as a limitation in studies with broadly homogenous samples. Participant demographics were not provided for one study (Kendall, Sloper, Lewin, & Parsons, 2003) so the authors were contacted, but were unable to provide this information. Many of the studies in Category B did not justify their choice of analytic approach or provided theoretical underpinnings for its use in the context of the research (Paper 1, Table 3), and most of the studies categorised as C and excluded were older studies or studies that were written in a different language and then translated into English, possibly having a detrimental effect on the methodology and content.
While most of the studies utilised purposive or convenience sampling, this was not always justified within the methodology, and resulted in limitations due to the restricted sample. The majority of studies used interviews or focus groups to collect their data, which are common methods to gain insight and understanding of a phenomenon (Gill, Stewart, Treasure, & Chadwick, 2008). The style of interviews was not always stated in studies which employed this method of data collection, and the differences in design may have affected the type and quality of data obtained. For example, more in-depth interviews may have explored one or two themes in detail, while those with more structure will have covered a pre-determined set of questions to explore a range of issues (Britten, 1995). Many of the Category B studies were older than the others in this review and appeared to lack the rigour of methodology, replicability, reporting trail and sensitivity to ethical concerns that the more recent studies made clear (Leung, 2015).

Part 2: Empirical Study

Sample and recruitment

I was fortunate to have the support of a local cardiology department with a database of families who could be contacted to participate in this research so that even though CHD is relatively rare in the population, I had access to many participants. Although all eligible families were contacted by the cardiology department staff, there was only an 8% response rate and so the sample may not be representative of their clinical population. Furthermore, not all parents who responded by post could be contacted by telephone or email, and so only those
who responded to communication were included in this study. It is not possible to compare this sample response size to similar studies because they are not explicit in their recruitment aims. IPA does not always have a target number of participants; rather data are collected until researchers reach theoretical saturation (Robinson, 2014). Smith (2015) recommends four to ten interviews, although basic elements for metathemes can occur within the first six interviews (Guest, Bunce, & Johnson, 2006). Furthermore, Sandelowski (1986) proposes that representativeness in qualitative research refers to the data, not the sample, and so it is up to the researcher to establish the typicality of the emerging themes from the participants’ reports. The sample in this study had a level of heterogeneity not always prominent in such studies; marital status included married, divorced, widowed, and living in different countries. Ethnicity was predominantly white but also included Asian and African descent and two of the children from the sample were born outside of the UK; however, this demographic information was not included in Tables 1 and 2 to reduce the possibility of participant identification.

**Ethical Considerations**

The NHS Research Ethics Committee (REC) expressed some concerns about the demographic information that parents were asked to provide, because there was a question about whether the child had a learning disability. It was felt that it was important to include the question, because 20% of children with CHD have a learning disability (Massin, Astadicko, & Dessy, 2007) which can add to carer burden (Datta, Russell, & Gopalakrishna, 2002), but to avoid distress for the parents, the question would be asked within the context of the statistic.
Confidentiality

Participants were notified in the PIS and prior to interview that all information gathered was confidential, with the exception of any disclosure of risk to self or other. Anonymity was maintained by assigning participant numbers and redacting names and locations from the transcripts. Audio recordings of interviews were deleted after transcription and all data were stored securely in line with data protection protocols.

Risk

It was anticipated that participants might find it helpful to share their views and provide feedback on their experiences; however, it was acknowledged that this could be difficult due to the personal and sensitive nature of the discussions. Steps were taken to minimise potential distress for participants, including reminding them that they did not have to answer questions they do not wish to. The researcher, a trainee clinical psychologist, used her therapeutic skills to respond empathically to verbal and non-verbal cues of distress. Parents were debriefed after the interview (see Appendix 14), and provided with contact details for support groups and paediatric psychologists they could contact if they wanted further information or support for any of the issues raised. A distress protocol was followed throughout the process (Appendix 15).

Methodology
There are a range of methodologies that can be used to develop an understanding of individuals’ experiences of phenomena. Initially, the researchers considered using thematic analysis (Braun & Clarke, 2006) to analyse the interview data because it would provide useful general themes that could be used to understand commonalities between parents of children with CHD. However, the research team believed that this approach might not have established a richer understanding of the individual’s experience and any unique insights that each case (or family) might bring (Smith, 2011). Semi-structured interviews allow for individual variation in the types of themes that are explored, although not all parents were asked the same questions which implies that some parents might have experienced problems which were not explored.

**Role of researcher and clinician**

My primary role during interviews was of researcher, although parents were aware that I was a trainee clinical psychologist with experience working clinically with families. In early interviews, it was a challenge to listen without offering summaries, reassurance and normalising, and this was evident when I transcribed the first three interviews. It became easier with later interviews because I encouraged parents to create a narrative with their own words, offering prompts only when necessary, and I developed the skill of switching from a clinician to a researcher. However, I felt that my clinical skills enabled me to be a more sensitive researcher, responding to their changing emotional state appropriately. Some of the parents I met asked questions about the emotional needs of their children and expressed a need for psychological guidance or support, knowing my role as a
clinical practitioner and my links with the paediatric psychology department. In these instances, I explained that I would like to discuss it further once the interview was finished as part of the debrief, and provided contact details for services who could provide support. One parent, who had not accessed support for their family, reflected that it had been a positive experience just speaking to someone who had an interest in and understanding of the condition, despite not being a parent or having experience of CHD in my family. I was touched by the parents’ openness in interviews, and felt it was important to record my personal thoughts in a reflective diary, not only to ensure reflexivity during analysis but also to discuss in supervision.

**Reflexivity**

Qualitative researchers bring their own preconceptions, expectations, knowledge and experience to the whole research process, so reflexivity is an important aspect of qualitative research (Smith et al. 2009). According to Richards & Emslie (2002) the professional background of a researcher can influence participant’s responses during interviews. The dynamic between participants who were parents of children with a health condition and the researcher who did not have children may have resulted in assumptions being made on both sides, such as understanding or lack thereof relating to parenting pressures. Conversely, due to the rarity of CHD, discussions could have been facilitated due to the researcher’s pre-existing knowledge of the condition meaning parents did not have to explain aspects of their experience as they may have to with a lay person.
It is important to consider the professional and personal background of researchers to ensure a rigorous approach to interpretation and analysis (Clancy, 2013). The background of the research team was briefly described, but this does not fully explain how researchers fitting into those particular demographics would reach the interpretative conclusions reported in the empirical paper. The interpretative process is likely to have been influenced by the researchers’ experiences, particularly whether they were themselves parents, as well as their clinical use of psychological models to understand experiences from a psychological perspective.

**Analysis**

I chose to transcribe all the interviews because this allowed me to immerse myself into the data at an early stage and pick out significant moments from the interviews that resonated. Transcribing interviews and re-reading transcripts for the analysis evoked emotions in me because I recalled the parent telling me about their experiences, and I felt a responsibility to communicate them within the write up. There were themes present in some of the transcripts that were not common across all of the interviews, including turning to faith as a support for the parents. For those who reported this, it was very important and I could see how it helped them through decision making and times of uncertainty. However, because this theme was only present in three interviews, it was not reported in the main paper. It was important to distance myself from the data and remind myself that I was not telling one parent’s story; I was trying to describe the parent experience.
Some of the parents’ accounts sounded more like monologues; they had much to say and it was so meaningful to them, and for some it was the first time they had really talked about their journey with CHD. At times it felt as a researcher that I was reducing their experience down to less than the sum of its parts by picking out emergent themes. I brought this issue to my supervisors’ attention and reflected on the purpose of the research being to explore the “parent experience”, not just one participant’s narrative.

**Clinical Implications**

Future research in this area has been mentioned in Paper 2. Research could also focus on the sibling experience of CHD because they would inevitably be impacted by the presence of CHD in their lives. Of particular interest would be a comparison of experiences when a sibling is older or younger than the child with CHD.

As a trainee clinical psychologist, I recognised areas where services could have stepped in to support the families, and provide education or respite at crucial times. Parents who received a postnatal diagnosis of CHD unfortunately missed out on the preparation that was available antenatally, and some were unaware of the support available in the early years, so there is certainly the need and opportunity to develop a pathway and support structure to ensure parents access services they need. As detailed in the discussion of both papers, transition points are key times when families are faced with additional, sometimes unexpected challenges, and so services should be mindful of these. The cardiology department in the hospital from which participants were recruited has addressed this already by offering “transition days” to bring families together for peer support when their child is
starting secondary school. The department was proactive in requesting this research because they understood the need to learn from parents what was helpful and what needed improving based on their experience. One of the questions in the interview referred to advice parents would give to others, and the responses could be shared with new parents without any analysis or interpretation, they are that powerful.

A further application of the findings of these papers would be to develop a parents’ guide to the CHD journey, so they have a resource full of the vast information that is too much to process in the early stages. This guide could contain details about specific diagnoses to avoid misuse of the internet (“Dr Google”) which can be unhelpful and anxiety provoking. It could also feature frequently asked questions and tips for parents when meeting with their consultant, similar to the strategies in Paper 1 and advice from parents in Paper 2. Basic information about the hospital, such as a map of departments and staff directory, could provide practical support to parents who live far away or have not visited before feel more familiar with the environment. It may be a useful resource to inform and educate families about how to communicate details of CHD to their child, when the time comes, and to acknowledge the emotional and mental health issues that can be associated with children with chronic health conditions. This psychologically informed approach could normalise their experiences.

Profile of Parents

Whilst it is acknowledged that every family has a unique experience and brings their own individual traits and resources to managing the challenge of parenting a
child with CHD, it can be useful to identify families who may be vulnerable and would benefit from additional support, whether this takes the form of psychological, social, financial or peer support.

The findings from the systematic review and empirical study generally showed that parents in a stable relationship who were able to draw on the support and understanding of a partner managed better than those whose partner was unavailable, whether due to geography, divorce or mental health difficulties. Parents who had children prior to the birth of their child with CHD had the advantage of knowing what behaviours were typical of a newborn so were more confident in seeking support when it was necessary, compared to first time parents who were uncertain of what was “normal” and so were hypervigilant to the slightest sign of distress. Having other children forced families to carry on with their lives, whereas an only child with CHD took the full attention and dedication of the parents, to the point of giving up work to care for them.

Parents who lived close to the hospital felt more secure in relation to the health care needs of their child. If they feared symptoms were emerging it was only a short trip to have them seen by their consultant. Most of the parents interviewed were in some form of employment and reported that their employers were supportive of taking time off when the child was in hospital for surgery. This flexibility enabled parents to retain some normality and maintain an income. For parents not in employment, their time was often devoted to their child and they could face financial difficulties if they were not supported by a partner who provided for the family.
Personal Reflections

Through meeting and interviewing the parents who participated, and immersing myself in the analysis, I was struck by the unwavering dedication they had for their families, in the face of so many adversities. Furthermore, despite the lack of understanding shown by people outside the family and lack of support provided by services, when the need is evident, parents still persevered, and in their own words, “just do what they have to for the sake of the child” with tenacity and resilience. This has included giving up work, opting not to have more children, moving to a completely new country and way of life, changing aspirations. It made me deeply reflect on the impact not only of having a child, but having a child who would be so dependent on a parent that it completely transformed their lives in a way unimaginable. I had great respect for the parents, and admiration that they used the opportunity of participating in research to support others who were about to embark on a similar journey.

Dissemination Plan

Papers 1 and 2 will be submitted for publication in academic journals. Findings will also be presented to the cardiology department at Alder Hey Hospital where the specialist psychology and surgical teams will be able to use them as evidence to inform clinical practice. In time, this could inform practice in other departments where common themes exist in other chronic childhood conditions. Finally, a lay summary of Paper 2 will be sent to participants to inform them of the findings and thank them for their contributions to the research.
References


Richards, H., & Emslie, C. (2000). The 'doctor'or the 'girl from the University'? Considering the influence of professional roles on qualitative interviewing. *Family practice, 17*(1), 71-75.


Appendices
Appendix 1

Author guidelines for the journal Pediatric Cardiology

The following information has been taken from guidance documentation accessed from: http://www.springer.com/medicine/cardiology/journal/246

Title Page
The title page should include:

- The name(s) of the author(s)
- A concise and informative title
- The affiliation(s) and address(es) of the author(s)
- The e-mail address, and telephone number(s) of the corresponding author
- If available, the 16-digit ORCID of the author(s)

Abstract
Please provide an abstract of 150 to 250 words. The abstract should not contain any undefined abbreviations or unspecified references.

Keywords
Please provide 4 to 6 keywords which can be used for indexing purposes.

Text Formatting
Manuscripts should be submitted in Word.

- Use a normal, plain font (e.g., 10-point Times Roman) for text.
- Use italics for emphasis.
- Use the automatic page numbering function to number the pages.
- Do not use field functions.
- Use tab stops or other commands for indents, not the space bar.
- Use the table function, not spreadsheets, to make tables.
- Save your file in docx format (Word 2007 or higher) or doc format (older Word versions).
References should be listed in the order they were cited in the text. Please note that we have recently changed the style of reference listing. Pediatric Cardiology no longer requests listing references in alphabetical order.

Citation
Reference citations in the text should be identified by numbers in square brackets. Some examples:
1. Negotiation research spans many disciplines [3].
2. This result was later contradicted by Becker and Seligman [5].
3. This effect has been widely studied [1-3, 7].

Reference list
The list of references should only include works that are cited in the text and that have been published or accepted for publication. Personal communications and unpublished works should only be mentioned in the text. Do not use footnotes or endnotes as a substitute for a reference list.
The entries in the list should be numbered consecutively.

Tables
- All tables are to be numbered using Arabic numerals.
- Tables should always be cited in text in consecutive numerical order.
- For each table, please supply a table caption (title) explaining the components of the table.
- Identify any previously published material by giving the original source in the form of a reference at the end of the table caption.
- Footnotes to tables should be indicated by superscript lower-case letters (or asterisks for significance values and other statistical data) and included beneath the table body.
Appendix 2
Noblit and Hare’s (1988) Seven-Step-Metasynthesis Approach

1. Deciding on a phenomenon
   A research question compatible with a qualitative approach was selected: Coping in parents of children with CHD.

2. Deciding on what qualitative studies are relevant to the research question
   Rigorous inclusion and exclusion criterion was created; which only included peer-reviewed studies, as their quality had been already assessed. After the search process was completed, all studies were assessed for relevance according to this criterion.

3. Becoming familiar with the studies
   The studies which were going to be included in the synthesis were read several times. Detailed demographic, methodological and findings data were extracted and tabulated from all 22 studies.

4. Determining how the studies related to each other
   The findings from the studies were carefully compared against each other to see if the data were directly comparable (reciprocal translation), in opposition (refutational) or represented a new line of enquiry. The themes were sufficiently similar to allow reciprocal translation, but attention was paid to studies which contained themes which stood out. Rather than subsume these themes they were identified and discussed.

5. The process of translation
   This stage involved a comparative scrutiny of themes and concepts between all the studies. I looked for similarities using hermeneutic intent and translated themes, using interpretative processes, from one study into the next and so on but was mindful to preserve the integrity and intricacies of the original data.

6. Synthesising translations
   Once the studies were translated into each other, the themes were clustered together according to their interpretative meaning and relationship to each other. Clustered themes were reconceptualised to formulate an over-arching umbrella theme which offered a new holistic interpretation.

7. Expressing the synthesis
   The resulting synthesis was expressed in written form which offered a comprehensive description and discussion of the themes.
### Appendix 3

**Example of Systematic Search Strategy**

<table>
<thead>
<tr>
<th>#</th>
<th>Query</th>
<th>Results</th>
</tr>
</thead>
<tbody>
<tr>
<td>S1</td>
<td>Congenital heart disease*</td>
<td>55146</td>
</tr>
<tr>
<td>S2</td>
<td>congenital heart defect*</td>
<td>64548</td>
</tr>
<tr>
<td>S3</td>
<td>congenital disorder</td>
<td>56734</td>
</tr>
<tr>
<td>S4</td>
<td>heart defect*</td>
<td>110393</td>
</tr>
<tr>
<td>S5</td>
<td>single ventricle</td>
<td>16983</td>
</tr>
<tr>
<td>S6</td>
<td>adjust*</td>
<td>1272282</td>
</tr>
<tr>
<td>S7</td>
<td>adapt*</td>
<td>2849516</td>
</tr>
<tr>
<td>S8</td>
<td>cope or coping</td>
<td>261844</td>
</tr>
<tr>
<td>S9</td>
<td>manage*</td>
<td>5451893</td>
</tr>
<tr>
<td>S10</td>
<td>deal</td>
<td>536699</td>
</tr>
<tr>
<td>S11</td>
<td>endure</td>
<td>12035</td>
</tr>
<tr>
<td>S12</td>
<td>withstand</td>
<td>40678</td>
</tr>
<tr>
<td>S13</td>
<td>family</td>
<td>2616143</td>
</tr>
<tr>
<td>S14</td>
<td>parent*</td>
<td>1113627</td>
</tr>
<tr>
<td>S15</td>
<td>Mother*</td>
<td>486326</td>
</tr>
<tr>
<td>S16</td>
<td>Father*</td>
<td>138858</td>
</tr>
<tr>
<td>S17</td>
<td>Mum*</td>
<td>168243</td>
</tr>
<tr>
<td>S18</td>
<td>Dad*</td>
<td>80335</td>
</tr>
<tr>
<td>S19</td>
<td>carer*</td>
<td>45091</td>
</tr>
<tr>
<td>S20</td>
<td>Caregiver*</td>
<td>129794</td>
</tr>
<tr>
<td>S21</td>
<td>famili*</td>
<td>1019985</td>
</tr>
<tr>
<td>S22</td>
<td>maternal</td>
<td>452309</td>
</tr>
<tr>
<td>S23</td>
<td>paternal</td>
<td>49918</td>
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<tr>
<td>S24</td>
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</tr>
<tr>
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<td>Foetus*</td>
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<tr>
<td>S30</td>
<td>son</td>
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<tr>
<td>S33</td>
<td>babies</td>
<td>149151</td>
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<tr>
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<td>newborn</td>
<td>753911</td>
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<tr>
<td>S35</td>
<td>toddler</td>
<td>23698</td>
</tr>
<tr>
<td>S36</td>
<td>S1 or S2 or S3 or S4 or S5</td>
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<td>S37</td>
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<tr>
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</tr>
<tr>
<td>S39</td>
<td>S24 or S25 or S26 or S27 or S28 or S29 or S30 or S31 or S32 or S33 or S34 or S35</td>
<td>6373344</td>
</tr>
<tr>
<td>S40</td>
<td>S36 AND S37 AND S38 AND S39</td>
<td>3455</td>
</tr>
<tr>
<td>S41</td>
<td>S40 AND qualitative</td>
<td><strong>90</strong></td>
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</table>
## Appendix 4
Walsh and Downe Quality Assessment Checklist

<table>
<thead>
<tr>
<th>Stage</th>
<th>Essential Criteria</th>
<th>Specific Prompts</th>
</tr>
</thead>
<tbody>
<tr>
<td>Scope and purpose</td>
<td>Clear statement of, and rationale for, research question/aims/purposes</td>
<td>Clarity of focus demonstrated</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Explicit purpose given, such as descriptive/explanatory intent, theory building, hypothesis testing</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Link between research and existing knowledge demonstrated</td>
</tr>
<tr>
<td></td>
<td>Study thoroughly contextualised by existing literature</td>
<td>Evidence of systematic approach to literature review, location of literature to contextualise the findings, or both</td>
</tr>
<tr>
<td>Design</td>
<td>Method/design apparent, and consistent with research intent</td>
<td>Rationale given for use of qualitative design</td>
</tr>
<tr>
<td></td>
<td>Discussion of epistemological/ontological grounding</td>
<td>Discussion of why particular method chosen is most appropriate/sensitive/relevant for research question/aims</td>
</tr>
<tr>
<td></td>
<td>Rationale explored for specific qualitative method (e.g. ethnography, grounded theory, phenomenology)</td>
<td></td>
</tr>
<tr>
<td>Sampling Strategy</td>
<td>Sample and sampling method appropriate</td>
<td>Selection criteria detailed, and description of how sampling was undertaken</td>
</tr>
<tr>
<td></td>
<td>Justification for sampling strategy given</td>
<td>Thickness of description likely to be achieved from sampling</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Any disparity between planned and actual sample explained</td>
</tr>
<tr>
<td>Analysis</td>
<td>Analytic approach appropriate</td>
<td>Approach made explicit (e.g. Thematic distillation, constant comparative method, grounded theory)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Was it appropriate for the qualitative method chosen?</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Was data managed by software package or by hand and why?</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Discussion of how coding systems/conceptual frameworks evolved</td>
</tr>
<tr>
<td></td>
<td></td>
<td>How was context of data retained during analysis</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Evidence that the subjective meanings of participants were portrayed</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Evidence of more than one researcher involved in stages if appropriate to epistemological/theoretical stance</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Did research participants have any involvement in analysis (e.g. member checking)</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Evidence provided that data reached saturation or discussion/rationale if it did not</td>
</tr>
<tr>
<td></td>
<td></td>
<td>Evidence that deviant data was sought, or discussion/rationale if it was not</td>
</tr>
<tr>
<td>Interpretation</td>
<td>Context described and taken account of in interpretation</td>
<td>Description of social/physical and interpersonal contexts of data collection Evidence that researcher spent time ‘dwelling with the data’, interrogating it for competing/alternative explanations of phenomena</td>
</tr>
<tr>
<td>----------------</td>
<td>----------------------------------------------------------</td>
<td>------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------------</td>
</tr>
<tr>
<td></td>
<td>Clear audit trail given</td>
<td>Sufficient discussion of research processes such that others can follow ‘decision trail’</td>
</tr>
<tr>
<td></td>
<td>Data used to support interpretation</td>
<td>Extensive use of field notes entries/verbatim interview quotes in discussion of findings Clear exposition of how interpretation led to conclusions</td>
</tr>
<tr>
<td>Reflexivity</td>
<td>Researcher reflexivity demonstrated</td>
<td>Discussion of relationship between researcher and participants during fieldwork Demonstration of researcher’s influence on stages of research process Evidence of self-awareness/insight Documentation of effects of the research on researcher Evidence of how problems/complications met were dealt with</td>
</tr>
<tr>
<td>Ethical</td>
<td>Demonstration of sensitivity to ethical concerns</td>
<td>Ethical committee approval granted Clear commitment to integrity, honesty, transparency, equality and mutual respect in relationships with participants Evidence of fair dealing with all research participants Recording of dilemmas met and how resolved in relation to ethical issues Documentation of how autonomy, consent, confidentiality, anonymity were managed</td>
</tr>
<tr>
<td>Relevance and Transferability</td>
<td>Relevance and transferability evident</td>
<td>Sufficient evidence for typicality specificity to be assessed Analysis interwoven with existing theories and other relevant explanatory literature drawn from similar settings and studies Discussion of how explanatory propositions/emergent theory may fit other contexts Limitations/weaknesses of study clearly outlined Clearly resonates with other knowledge and experience Results/conclusions obviously supported by evidence Interpretation plausible and ‘makes sense’ Provides new insights and increases understanding Significance for current policy and practice outlined Assessment of value/empowerment for participants Outlines further directions for investigation Comment on whether aims/purposes of research were achieved</td>
</tr>
</tbody>
</table>
Appendix 5
Quality Assessment of Excluded Papers

<table>
<thead>
<tr>
<th></th>
<th></th>
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</tr>
</thead>
<tbody>
<tr>
<td>1: Clear statement of, and rationale for, research question/aims/purposes</td>
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<td>0.5</td>
<td>0.5</td>
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</tr>
<tr>
<td>2: Study thoroughly contextualised by existing literature</td>
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<td>1</td>
<td>1</td>
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<td>0.5</td>
</tr>
<tr>
<td>3: Method/design apparent, and consistent with research intent</td>
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<td>0</td>
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<tr>
<td>4: Data collection strategy apparent and appropriate</td>
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<td>1</td>
</tr>
<tr>
<td>5: Sample and sampling method appropriate</td>
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<td>0</td>
<td>0</td>
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<td>0.5</td>
</tr>
<tr>
<td>6: Analytic approach appropriate</td>
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<td>0.5</td>
<td>0</td>
<td>0.5</td>
<td>0.5</td>
</tr>
<tr>
<td>7: Context described and taken account of in interpretation</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>8: Clear audit trail given</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>9: Data used to support interpretation</td>
<td>1</td>
<td>0.5</td>
<td>0</td>
<td>1</td>
<td>1</td>
</tr>
<tr>
<td>10: Researcher reflexivity demonstrated</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
<td>0</td>
</tr>
<tr>
<td>11: Demonstration of sensitivity to ethical concerns</td>
<td>0.5</td>
<td>0</td>
<td>0</td>
<td>1</td>
<td>0</td>
</tr>
<tr>
<td>12: Relevance and transferability evident</td>
<td>0.5</td>
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</tr>
<tr>
<td>Total (category)</td>
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<td>5.5 (C)</td>
<td>2 (C)</td>
<td>5.5 (C)</td>
<td>5 (C)</td>
</tr>
</tbody>
</table>

Maximum quality rating score is 12. These studies fell into category C (0-5.5).
Appendix 6

Author Guidelines for Congenital Heart Disease

The following information has been taken from guidance documentation accessed from:


Manuscripts must be submitted in .doc or .rtf file format. Make sure all text is double-spaced. Please be sure all text, tables, and figures are clear and readable in the system-rendered PDF and HTML views. Be sure to keep a back up copy of the file for reference, as accepted manuscripts are not returned.

There are no restrictions on the length of any article type. If the article is of interest to the editors but is deemed to be too long, cuts will be requested in a revision.

Title Page

The title page should be the first page of the manuscript text document and contain all the following:

- Title
- Full names and affiliations for all authors, including the highest academic degree
- Full postal address, telephone number, fax number, and e-mail address for the corresponding author, to whom the proofs will be sent
- Conflict of Interest statement
- Disclosure of grants or other funding

Abstracts and Keywords

The abstract, on the page following the title page, is required for all papers except a Letter to the Editor and must be 300 words or less. The abstracts for Reviews and Commentaries should be unstructured. All other articles requiring an abstract should submit a structured abstract using the following headings, as appropriate: Objective, Design, Setting, Patients, Interventions, Outcome Measures, Results, and Conclusions (JAMA 1992;267:42–44). Up to six key words suitable for indexing must be provided with the abstract.

Body Text

Research papers should be structured as follows: Title page, as above; Abstract and keywords; Introduction; Methods; Results; Discussion; Acknowledgments (optional); Author Contributions; References; Tables; Figure legends (double-spaced); Figures.

illustrations, and tables. Start each of these sections on a new page, numbered consecutively in the upper right-hand corner, beginning with the title page.

**References**

References for Congenital Heart Disease should follow the American Medical Medical Association Manual of style system.

**Tables**

All tables should be double-spaced, and the text should be clear and readable for review. Title all tables at the top, and number them in order of their citation in the text. Any notes should appear at the bottom of the table. For review, tables may be embedded in the flow of text as they are mentioned, position at the end of the text, or submitted in a separate file. For accepted papers, tables must be submitted separately from the main manuscript document, in a text file (.doc or .rtf), with each table beginning on a new page.
Appendix 7
Ethical approval

North West - Greater Manchester Central Research Ethics Committee

Please note: This is the favourable opinion of the REC only and does not allow you to start your study at NHS sites in England until you receive HRA Approval

07 April 2017
Miss Midori Lumsden
Trainee Clinical Psychologist
University of Manchester
Zochonis Building
Brunswick Street
Manchester
M13 9PL

Dear Miss Lumsden

Study title: Children with Single Ventricle Congenital Heart Defects; considering the parent experience
REC reference: 17/NW/0165
Protocol number: 1
IRAS project ID: 212134

Thank you for your correspondence of 5th April 2017, responding to the Committee’s request for further information on the above research and submitting revised documentation.

The further information has been considered on behalf of the Committee by the Chair.

We plan to publish your research summary wording for the above study on the HRA website, together with your contact details. Publication will be no earlier than three months from the date of this opinion letter. Should you wish to provide a substitute contact point, require further information, or wish to make a request to postpone publication, please contact hra.studyregistration@nhs.net outlining the reasons for your request.
Confirmation of ethical opinion

On behalf of the Committee, I am pleased to confirm a favourable ethical opinion for the above research on the basis described in the application form, protocol and supporting documentation as revised, subject to the conditions specified below.

Conditions of the favourable opinion

The REC favourable opinion is subject to the following conditions being met prior to the start of the study.

Management permission must be obtained from each host organisation prior to the start of the study at the site concerned.

Management permission should be sought from all NHS organisations involved in the study in accordance with NHS research governance arrangements. Each NHS organisation must confirm through the signing of agreements and/or other documents that it has given permission for the research to proceed (except where explicitly specified otherwise).


Where a NHS organisation’s role in the study is limited to identifying and referring potential participants to research sites (“participant identification centre”), guidance should be sought from the R&D office on the information it requires to give permission for this activity.

For non-NHS sites, site management permission should be obtained in accordance with the procedures of the relevant host organisation.

Sponsors are not required to notify the Committee of management permissions from host organisations.

Registration of Clinical Trials

All clinical trials (defined as the first four categories on the IRAS filter page) must be registered on a publicly accessible database within 6 weeks of recruitment of the first participant (for medical device studies, within the timeline determined by the current registration and publication trees).

There is no requirement to separately notify the REC but you should do so at the earliest opportunity e.g. when submitting an amendment. We will audit the registration details as part of the annual progress reporting process.

To ensure transparency in research, we strongly recommend that all research is registered but for non-clinical trials this is not currently mandatory.

If a sponsor wishes to request a deferral for study registration within the required timeframe, they should contact hra.studyregistration@nhs.net. The expectation is that all clinical trials will
be registered, however, in exceptional circumstances non registration may be permissible with prior agreement from the HRA. Guidance on where to register is provided on the HRA website.

It is the responsibility of the sponsor to ensure that all the conditions are complied with before the start of the study or its initiation at a particular site (as applicable).

Ethical review of research sites

NHS sites

The favourable opinion applies to all NHS sites taking part in the study, subject to management permission being obtained from the NHS/HSC R&D office prior to the start of the study (see "Conditions of the favourable opinion" below).

Non-NHS sites

Approved documents

The final list of documents reviewed and approved by the Committee is as follows:

<table>
<thead>
<tr>
<th>Document</th>
<th>Version</th>
<th>Date</th>
</tr>
</thead>
<tbody>
<tr>
<td>Contract/Study Agreement [Research contract]</td>
<td>1</td>
<td>06 April 2016</td>
</tr>
<tr>
<td>Evidence of Sponsor Insurance or indemnity (non NHS Sponsors only) [Insurance assessment form]</td>
<td>1</td>
<td>13 January 2017</td>
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<tr>
<td>Interview schedules or topic guides for participants [Interview Schedule]</td>
<td>3</td>
<td>22 December 2016</td>
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<tr>
<td>IRAS Application Form [IRAS_Form_21022017]</td>
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<td>21 February 2017</td>
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<tr>
<td>IRAS Checklist XML [Checklist_21022017]</td>
<td></td>
<td>21 February 2017</td>
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<tr>
<td>Letter from sponsor [Letter from sponsor]</td>
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<td>10 February 2017</td>
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<tr>
<td>Letters of invitation to participant [Updated letter of invitation]</td>
<td>1</td>
<td>20 February 2017</td>
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<tr>
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<tr>
<td>Other [Risk Assessment]</td>
<td></td>
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<tr>
<td>Other [Lone Working Policy]</td>
<td>1</td>
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<tr>
<td>Other [Thank you letter wording]</td>
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<td>10 February 2017</td>
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<tr>
<td>Other [Distress Protocol]</td>
<td>2</td>
<td>04 April 2017</td>
</tr>
<tr>
<td>Participant consent form [Participant Consent to Contact Form]</td>
<td>1</td>
<td>22 December 2016</td>
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<tr>
<td>Participant consent form</td>
<td>3</td>
<td>05 April 2017</td>
</tr>
<tr>
<td>Participant information sheet (PIS) [Participant Debrief Sheet]</td>
<td>1</td>
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<tr>
<td>Participant information sheet (PIS) [Participant Information Sheet]</td>
<td>2</td>
<td>28 March 2017</td>
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<tr>
<td>Research protocol or project proposal [Research Protocol]</td>
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<td>04 January 2017</td>
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<tr>
<td>Summary CV for Chief Investigator (CI) [M Lumsden Research CV]</td>
<td>1</td>
<td>03 January 2017</td>
</tr>
<tr>
<td>Summary CV for student [M Lumsden Research CV]</td>
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<td>03 January 2017</td>
</tr>
<tr>
<td>Summary CV for supervisor (student research) [A Wittkowski Research CV]</td>
<td>2</td>
<td>22 January 2017</td>
</tr>
<tr>
<td>Summary CV for supervisor (student research) [D Smith Research CV]</td>
<td>2</td>
<td>19 September 2016</td>
</tr>
</tbody>
</table>
Summary, synopsis or diagram (flowchart) of protocol in non-technical language [Study Protocol]

Statement of compliance

The Committee is constituted in accordance with the Governance Arrangements for Research Ethics Committees and complies fully with the Standard Operating Procedures for Research Ethics Committees in the UK.

After ethical review

Reporting requirements

The attached document “After ethical review – guidance for researchers” gives detailed guidance on reporting requirements for studies with a favourable opinion, including:

- Notifying substantial amendments
- Adding new sites and investigators
- Notification of serious breaches of the protocol
- Progress and safety reports
- Notifying the end of the study

The HRA website also provides guidance on these topics, which is updated in the light of changes in reporting requirements or procedures.

User Feedback

The Health Research Authority is continually striving to provide a high quality service to all applicants and sponsors. You are invited to give your view of the service you have received and the application procedure. If you wish to make your views known please use the feedback form available on the HRA website:
http://www.hra.nhs.uk/about-the-hra/governance/quality-assurance/

HRA Training

We are pleased to welcome researchers and R&D staff at our training days – see details at
http://www.hra.nhs.uk/hra-training/

17/NW/0165 Please quote this number on all correspondence

With the Committee’s best wishes for the success of this project.

Yours sincerely

[Signature]

Signed on behalf of the Chair, Dr Barbara Potrata

Email:nrescommittee.northwest-gmoentral@nhs.net
Miss Midori Lumsden
Trainee Clinical Psychologist
University of Manchester
Zochonis Building
Brunswick Street
Manchester
M13 9PL

07 April 2017

Dear Miss Lumsden,

Letter of HRA Approval

Study title: Children with Single Venticle Congenital Heart Defects; considering the parent experience
IRAS project ID: 212134
Protocol number: 1
REC reference: 17/NW/0165
Sponsor University of Manchester

I am pleased to confirm that HRA Approval has been given for the above referenced study, on the basis described in the application form, protocol, supporting documentation and any clarifications noted in this letter.

Participation of NHS Organisations in England
The sponsor should now provide a copy of this letter to all participating NHS organisations in England.

Appendix B provides important information for sponsors and participating NHS organisations in England for arranging and confirming capacity and capability. Please read Appendix B carefully, in particular the following sections:

- Participating NHS organisations in England – this clarifies the types of participating organisations in the study and whether or not all organisations will be undertaking the same activities.
- Confirmation of capacity and capability - this confirms whether or not each type of participating NHS organisation in England is expected to give formal confirmation of capacity and capability. Where formal confirmation is not expected, the section also provides details on the time limit given to participating organisations to opt out of the study, or request additional time, before their participation is assumed.
- Allocation of responsibilities and rights are agreed and documented (4.1 of HRA assessment criteria) - this provides detail on the form of agreement to be used in the study to confirm capacity and capability, where applicable.

Further information on funding, HR processes, and compliance with HRA criteria and standards is also provided.
Appendix 8
Invitation letter
Dear parent/guardian(s)

The cardiology team at Alder Hey are collaborating with a trainee clinical psychologist, Midori Lumsden, who is completing her doctoral training at the University of Manchester. As part of her studies, she is completing a research project to explore the experiences of parents of children with a single ventricle congenital heart defect.

You have been identified as a potential participant by the care team at Alder Hey Children’s Hospital as your child has had their Fontan surgery at least six months ago. Please note that no confidential or personal information has been shared with the trainee or the university. We are sending this letter out on behalf of Midori Lumsden.

In this pack, you will find information about the research project, and what it would involve should you decide to participate. If you would like more information, or just to talk about the project, please contact Midori Lumsden directly by phone, or by email: details are provided on the information sheet.

If you wish to participate in the project, please return the reply slip in the envelope provided with your contact details and Midori Lumsden will contact you to arrange a meeting.

Yours sincerely

Dr Heather Smith & Dr Emma Twigg
Clinical Psychologist Consultant Psychologist
Psychological Services, Alder Hey Children’s Hospital
Appendix 9
Participant Information Sheet
Participant Information Sheet

Children with Single Ventricle Congenital Heart Defects; considering the parent experience

Chief Investigator: Midori Lumsden

We would like to invite you to take part in a research study. Before you decide, we would like you to understand why the research is being done and what it would involve for you. Please read the following information carefully and discuss it with others if you wish. If anything seems unclear or if you would like more information, contact us at any time. Take time to decide whether or not you wish to take part.

What is the purpose of the study?
We are carrying out this study in order to understand the experience of parents who have a child with a single ventricle congenital heart defect, from diagnosis to post-surgery. We are particularly interested in finding out what parents need, and what makes it easier or harder to meet these needs. This will help the cardiology service improve so they can offer the most useful support at the right time.

Why have I been invited?
You have been invited to take part in this study because your child has had their Fontan stage of surgery at least 6 months ago. We are looking for 8-10 parents whose child has had their surgery and is still open to the cardiology team at Alder Hey Children’s Hospital.

Do I have to take part?
No, you do not have to participate in this study if you do not want to. Participation is completely voluntary. If you decide to take part, you will be given this information sheet to keep and then you will be asked to sign a consent form (a copy of which you will be given to keep). If you decide to participate, but change your mind later, you are free to withdraw at any point during the study, without giving a reason. Your decision to withdraw from the study or a decision not to take part will not affect your legal rights or your child’s medical care, and you will not be disadvantaged in any way.

What will happen if I decide to take part?
If you would like to take part in this study, we will arrange a convenient time to meet with you to tell you more about the study and to answer any questions you may have. The interview can take place at either your home address, a local GP surgery, Alder Hey Children’s Hospital or a place of your choice that is convenient to you and your family, whichever you prefer.

What does this study involve?
Participation will involve meeting with a researcher for around 1-2 hours to talk about your experiences as a parent since your child received their diagnosis. This will take the form of a semi-structured interview, and will involve being asked a number of open-ended questions allowing you to share your thoughts. Questions include “Tell me about when you received the diagnosis of your child’s heart problem”, “What effect has your child’s heart problem had on you as parents?”, “How has it affected you as a family?” and “What was your experience of services or people that were there to support you?” as well as prompts to explore these areas further.
You will also be asked to provide some brief details, such as diagnostic information and demographics relating to your family to provide context and help us understand where your experience and perspective is coming from. The information you provide will be anonymised.

What are the possible benefits of taking part?
Many people find it helpful to share their views and want to provide some feedback about their experiences. Although we don’t expect any direct benefit of taking part in this interview, the information we collect may help improve the support given to parents, families and children in the future.

**What are the possible disadvantages and risks of taking part?**
For some people, talking about their experiences and sharing their views can be difficult because it can be sensitive and personal. There is a possibility that talking about your experience may bring up strong emotions for some people. The researcher will be aware of this possibility and proceed only if you wish to do so. She will also remind you can withdraw from the study at any time. One other disadvantage of participating in this study is the time commitment of taking part in an interview for up to 2 hours.

**Will my taking part in the study be kept confidential?**
The interview will be recorded using an audio-recording device. It will then be typed up by the researcher who interviewed you, at which point the information will be anonymised. This means that your name and child’s identity will be taken out, and an ID code will be used instead. All data will be stored securely. For monitoring and auditing purposes, study data and materials may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust. All will have a duty of confidentiality to you as a participant. Personal data such as names and addresses recorded for purposes of contact will be destroyed at the end of the study, however consent forms are kept as essential documents. Information will remain strictly confidential and will not be shared outside the research team. There is one exception to this: If you tell us anything that makes us think that you, your child, or anyone else is at risk of harm, we will have to share this information, for example, with your consultant at Alder Hey or your GP.

When we publish the findings of this study, we may use direct quotes, but these will be used in such a way that they will not reveal your identity. Written transcripts of the interviews will be kept for a minimum of 5 years after the date of any publication which is based upon it, to follow recommended good practice guidelines for research.

**What will happen if I do not want to carry on with the study?**
You can withdraw from the study completely at any time without giving a reason and without any consequence to your child’s current or future NHS treatment. You can withdraw from the study, even if you have begun your interview. No further data will be collected from the moment you withdraw. Data provided until that point may be retained and anonymised. If the researcher has already begun to analyse collected data, it may not be possible to distinguish and separate this from other participants’ data.

**What will happen to the results of this study?**
Once all the information has been collected and analysed, the findings will be submitted for publication in an academic journal. A summary of findings will be sent to all participants by post. We will also share the results with other families, health care professionals and researchers at conferences. In all cases, any information you provided will be anonymous and used in such a way so they will not identify a particular participant.

**Who is organising the study?**
This study has been requested by the Cardiology Department at Alder Hey Children’s NHS Foundation Trust, and the clinical psychologists working within the cardiology service. The study is being undertaken by a Trainee Clinical Psychologist, Midori Lumsden, as part of a Doctorate in Clinical Psychology at the University of Manchester. This study is supervised by Dr Anja
Wittkowski, Senior Lecturer in Clinical Psychology, who is based at the University of Manchester and also works for the Greater Manchester Mental Health Trust.

**What if there is a problem (complaint)?**

If you have a minor complaint then please contact the researcher(s) in the first instance: Midori Lumsden – midori.lumsden@postgrad.manchester.ac.uk, Anja Wittkowski – anja.wittkowsi@manchester.ac.uk.

If you wish to make a formal complaint or if you are not satisfied with the response you have gained from the researchers in the first instance, then please contact the Research Governance and Integrity Manager, Research Office, Christie Building, University of Manchester, Oxford Road, Manchester, M13 9PL, by emailing: research.complaints@manchester.ac.uk or by telephoning 0161 275 2674 or 275 2046.

In the unlikely event that something does go wrong and you are harmed during the study, you may have grounds for a legal action for compensation against the University of Manchester or NHS Trust but you may have to pay your legal costs. The routine NHS complaints mechanisms will still be available to you, and PALS (Patient Advice and Liaison Service) should be contacted if you have a complaint about the research you wish to raise with Alder Hey Children’s Hospital (0151 252 5374 or 0151 252 5161).

**Who has reviewed the study?**

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed by North West-Greater Manchester South Research Ethics Committee and was given a favourable opinion on 7th April 2017. The REC reference number is 17/NW/0165.

*Thank you for considering taking part in this study.*

**Contacts for further information**

Midori Lumsden  
Chief Investigator for this study  
The University of Manchester, School of Health Sciences, Division of Psychology and Mental Health,  
2nd Floor Zochonis Building, Brunswick Street  
Manchester M13 9PL  
Tel. 07732 058528  
Email: Midori.lumsden@postgrad.manchester.ac.uk

Dr Anja Wittkowski  
Academic Supervisor for this study  
The University of Manchester, School of Health Sciences, Division of Psychology and Mental Health,  
2nd Floor Zochonis Building, Brunswick Street  
Manchester M13 9PL  
Tel. 0161 306 0400  
Email: anja.wittkowsi@manchester.ac.uk

Dr Emma Twigg and Dr Heather Smith  
Consultant Clinical Psychologist / Clinical Psychologist  
Psychological Services (Paediatrics)  
Alder Hey Children’s Hospital NHS Foundation Trust  
Building No. 1, Interim Estate,  
Eaton Road, Liverpool  
L12 2AP  
Tel: 0151 252 5586
Appendix 10
Participant consent to contact form
Consent to Contact Form

Study Title: Children with Single Ventricle Congenital Heart Defects; considering the parent experience

Chief Investigator: Midori Lumsden

Thank you for showing interest in the above study which is described more fully in the Participant Information Sheet. If you are interested in taking part in this study and would like the researcher to contact you, please give your details below. You should only provide the information if you are happy to be contacted in that way. For example, if you do not want to be contacted by phone, then do not provide a phone number.

Please note the following points in relation to the processing of your data:
- Data will be held securely by the research team on behalf of the University of Manchester according to the University’s data protection and information security policies.
- Access to the data will be restricted to the research team for the sole purpose of contacting you about this study.
- Your data will not be shared with any third party without your written permission.
- The details collected will only be stored for as long as required to find out if you wish to take part in the study. Once no longer needed, that data will be destroyed securely.
- If you decide to change your mind about being contacted about the study or would like your details to be destroyed you can contact Midori Lumsden (Chief investigator) on 07732 058528.

Once you have completed your details, please ensure that you have added your signature then tear the bottom half off and post it back in the envelope provided, or give it to your health care professional to return to the research team.

I am happy to provide my personal details so that I can be contacted about this study.

<table>
<thead>
<tr>
<th>Parent’s name</th>
<th></th>
</tr>
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<tbody>
<tr>
<td>Signature</td>
<td></td>
</tr>
<tr>
<td>Today’s date</td>
<td></td>
</tr>
</tbody>
</table>

Please complete the details below:

<table>
<thead>
<tr>
<th>Contact by letter</th>
<th>Address</th>
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</tbody>
</table>

<table>
<thead>
<tr>
<th>Contact by phone</th>
<th>Preferred contact number</th>
</tr>
</thead>
<tbody>
<tr>
<td></td>
<td>When would you prefer to be contacted? (please circle)</td>
</tr>
<tr>
<td></td>
<td>Morning/ Afternoon/ Evening/ Don’t Mind</td>
</tr>
<tr>
<td></td>
<td>Can a voicemail message be left on this telephone number? (please circle)</td>
</tr>
<tr>
<td></td>
<td>Yes / No</td>
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</table>

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<tr>
<th>Contact by email</th>
<th>Email address</th>
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</thead>
</table>
Appendix 11
Consent to participate form
CONSENT FORM

Title of Project: Children with Single Ventricle Congenital Heart Defects; considering the parent experience
Name of Researcher: Midori Lumsden

University of Manchester in collaboration with Alder Hey Children’s NHS Foundation Trust
Department of Cardiology

<table>
<thead>
<tr>
<th>Please read</th>
<th>Please initial box</th>
</tr>
</thead>
<tbody>
<tr>
<td>1</td>
<td>I confirm that I have read the information sheet dated....................... (version.........) for the above study. I have had the opportunity to consider the information, ask questions and have had these answered satisfactorily.</td>
</tr>
<tr>
<td>2</td>
<td>I understand that my participation is voluntary and that I am free to withdraw at any time without giving any reason, without my child’s medical care or our legal rights being affected, up until the research data have been analysed.</td>
</tr>
<tr>
<td>3</td>
<td>I understand that data collected during the study may be looked at by individuals from the University of Manchester, from regulatory authorities or from the NHS Trust, where it is relevant to my taking part in this research. I give permission for these individuals to have access to my data.</td>
</tr>
<tr>
<td>4</td>
<td>I give permission for direct quotes to be used in the write up of this research. I understand that these will be anonymous and no identifiable information will be used.</td>
</tr>
<tr>
<td>5</td>
<td>I consent to the research interview being audio recorded.</td>
</tr>
<tr>
<td>6</td>
<td>I consent to the audio recordings to be transcribed, with any names/places anonymised.</td>
</tr>
<tr>
<td>7</td>
<td>I agree to take part in the above study.</td>
</tr>
</tbody>
</table>

I confirm that I understand what the study involves and am willing to participate. I consent to the above points.

Signed:........................................... Researcher signature: ...........................................

Name: ........................................... Researcher name: ...........................................

Date: ........................................... Date: ...........................................

When completed: 1 copy for participant; 1 original for researcher site file.
Appendix 12
Demographic sheet
Children with SVCHD; considering the parent experience

Demographic information sheet

Participant number:

Age at interview:

Gender:

Ethnicity:

Current age of child:

Child’s specific diagnosis/diagnoses:

Current health of child (well/variable/unwell):

Any diagnosed Learning Disability:

Number of other children in family and family composition:

Health of other children:

Relationship status:

Employment status:
Appendix 13
Interview schedule
Children with Single Ventricle Congenital Heart Defects; considering the parent experience

Topic Guide for semi structured interview

Before you start:

Thank the participant for agreeing to be interviewed. As you are aware I am a trainee clinical psychologist conducting interviews for my doctoral research project.

The purpose of this research is to explore the experiences of parents who have a child who has been diagnosed with single ventricle congenital heart disease, who has completed their surgical procedures.

Everything you say in this interview will be anonymous and your confidentiality maintained

Are you still OK for us to tape record this conversation?

Please remember that it is OK to stop at any point, or refuse to answer any questions during this interview

Do you have any concerns before we start?

There are four sections to this interview: opening, your experience, support and closing statement.

What follows is a guide. The order and exact content of the questions will be determined by the participant so the order of the questions may vary as the interview develops. Probe and ask for examples as the time permits.

Opening statement

1. Tell me about when you received the diagnosis of <child’s name> heart problem

2. How have things been since this time?

3. Tell me what it was like when <child’s name> was growing up.

Prompts: Could you tell me more about that? In what way? What else can you tell me about that? When they had their surgery? As they reached milestones? When they went to school?

Your experience

4. What effect has your child’s heart problem had on you as parents?

5. How has it affected you as a family?

6. What is the relationship like with your child?
Prompts: Could you tell me more about that? In what way? What else can you tell me about that? What makes it easier? What makes it harder?

Support

7. What was your experience of services or people that were there to support you?

Prompts: What advice could you offer to other parents that they might find helpful? Things they’ve done themselves? Things others have done for them? Why would this help other parents? How would this be delivered?

Closing statement

8. Is there anything else we haven’t covered already about your experience that you would like to talk about before we finish?

Prompts: Could you tell me more about that? In what way? What else can you tell me about that?

Following interview

Thank participant.

Ask participants how they have found the interview.

Clarify whether participant has experienced any distress above what might be expected (due to discussing sensitive issues).

Researcher to give participant contact details of support organisations and signpost participant to appropriate agencies if they are distressed.
Appendix 14
Participant debrief sheet
Children with SVCHD; considering the parent experience  
Participant Debrief Sheet

Thank you for participating in this research. We hope you found it interesting. However, if you have found any part of this experience distressing, there are a number of people and organisations available for support.

- If you would like to speak to one of the researchers, please contact us on 07732 058528 or by email on midori.lumsden@postgrad.manchester.ac.uk. Alternatively, you can contact Dr Anja Wittkowski by writing to University of Manchester, Zochonis Building, Brunswick Street, Manchester, M13 9PL or emailing anja.wittkowski@manchester.ac.uk.
- If you feel as though you are struggling to cope, or feeling low in mood, it is important that you go to your GP to ask for support.
- You can also talk to the cardiac liaison nurses (0151 252 5291) or the psychologists working in the cardiology team at Alder Hey Children’s Hospital (0151 252 5586 – Emma Twigg or Heather Smith) who will be able to signpost you to an appropriate support service.
- There are also a number of charitable organisations and support networks listed below that you can contact.

<table>
<thead>
<tr>
<th>Organisations</th>
</tr>
</thead>
<tbody>
<tr>
<td><strong>Little Hearts Matter</strong></td>
</tr>
<tr>
<td>0121 455 8982</td>
</tr>
<tr>
<td><a href="mailto:info@lhm.org.uk">info@lhm.org.uk</a></td>
</tr>
<tr>
<td><a href="https://www.lhm.org.uk">https://www.lhm.org.uk</a></td>
</tr>
<tr>
<td>From initial antenatal diagnosis, through treatments and into life at home, Little Hearts Matter works with children, young adults and families to reduce the isolation, fear and lack of understanding created when a child is diagnosed as having half a working heart.</td>
</tr>
</tbody>
</table>

| **Children’s Heart Association**                   |
| 01706 221988                                       |
| information@heartchild.info                        |
| http://heartchild.info/web                         |
| Based in the North West of England, the Children’s Heart Association offers support and local unit information to the families of children treated at Alder Hey and Manchester Children’s Hospital. |

| **Contact a Family**                               |
| 0808 808 3555                                      |
| helpline@cafamily.org.uk                          |
| http://www.cafamily.org.uk                        |
| Contact a Family offer information, support and advice for parents of children with medical conditions and disabilities. They have a range of guidance for parents online. |

| **NHS Direct**                                     |
| 111                                                |
| Open 24 hours a day. They provide health advice and information. |

| **Samaritans**                                     |
| 0845 7909090                                       |
| Open 24 hours a day. They offer confidential emotional support by telephone, email, text, letter and face to face. |
Appendix 15
Distress protocol
Children with Single Ventricle Congenital Heart Defects; Considering the parent experience

Distress Management Protocol

**During recruitment, the researcher will:**
- Remind participants of the potential risks of taking part in the study, as outlined in the participant information sheet.
- Follow procedures described below if a participant wishes to stop or continue.

**If a participant appears to become distressed, the researcher will:**
- Encourage the participant to take a break from answering questions.
- Acknowledge that talking about their experiences can be distressing.
- Offer support by reassuring participants that they do not need to answer a question(s) if they do not wish to.
- Ask if they would like to stop the interview or would like to continue.

**If the participant would like to stop:**
- Finish the interview at that point and offer to return at another time.
- If the participant withdraws their consent to participate in the study, then another time will not be arranged, and no further data will be collected.

**If the participant would like to continue:**
- Take time at the end of the assessment process or the interview to talk informally, and encourage the participant to access further support depending on their level of distress (e.g. assess the level of distress as low, moderate or severe).
  - If the participant is still under the care of the paediatric psychology department, then discuss with the participant if they want their clinical team to be made aware of their feelings in order to gain additional support from staff. Discuss if the participant would like to make the psychology team aware of their distress themselves or if they consent for the researcher to do so.
  - If the participant is no longer open to the paediatric psychology department, then discuss with the participant if they want their current health care professional (e.g. GP) to be made aware of their feelings in order to gain additional support from professionals. Discuss if the participant would like to make that professional aware of their distress themselves or if they consent for the researcher to do so.
- If the participant has any questions or requires some reassurance about the research at a future point, they should be encouraged to contact the chief investigator using the contact details given on the participant information sheet.
- If the participant still appears to be distressed when the assessment process or the interview is over, then the researcher will offer to phone back in a couple of days to ensure the distress has not escalated and to reiterate the sources of support. A debrief sheet with additional information can also be left with the participant.
- If the participant’s distress is severe enough to increase the risk to their safety they will be advised to contact their health care provider, crisis team, The Samaritans, or their local Accident & Emergency department if necessary.

If there is a concern about the participant’s level of distress, the researcher will complete an incident form which will be shared with the field supervisor and the project team.
Appendix 16
Excerpt of transcription with descriptive and conceptual comments

P: So I was looking after this child as you should, but I wasn’t upset, to the point that I should be | remember when he was, when he was first taken into special care, grrr, there wasn’t really anything wrong with him but they said that they just wanted to check a few things, and I remember crying | and the midwife wrote on my notes “query post-natal depression, mum upset”. Well, no, it’s just a bit emotional, I’m like 6 hours out of labour and you’ve got a bit of a query! But then after that I was fine, absolutely fine, and the child grrr, went into the ward. [WHARD], and then he collapsed again the next day, so we ended up on ICU, and they decided to do surgery, but they couldn’t get a bed, and then he collapsed again, so they did surgery quicker than they’d planned, and it was all just happening and it was like, well yeah this is, this is what you do, you sit there with your baby and you do it, it was fine. And then after he’d had his surgery – so he had his surgery at 10 days, he had his chest open for three and a half weeks, so we were on ICU for four weeks, and then then he | cleaned the drugs wrong and things and he collapsed again and | Oh my goodness | P: So it all went on for like six months | and after about three months, yeah after about three months, I was like | we were doing alternate nights, sleeping with *child*, and one of us in [parent residential unit] or going home, and I just went home. I was like “I don’t want this anymore, I don’t want anything to do with my son, I don’t want anything to do with my husband!“ [laughs] And I just went, I was like, that’s it | Wow | P: Errr, cos we hadn’t had any support at this point really, nobody really, you know, even the cardiac liaison nurses hadn’t become involved and we didn’t know anybody to help us, it was just other parents, and I went. It was quite funny really, cos I went and it was a freezing cold night, so I put all the heaters, fan heaters on in the house, and put whatever, everything electrical on in the house, the belly and everything, and *father* was ringing me. And the phone wasn’t working cos the electricity had gone off, so I wasn’t answering the phone. And in the end, the next door neighbour knocked on the door [laughs] she was white as a sheet, she went [firmly] “phone your husband now!” and I phoned *father* and he thought I’d committed suicide! [Cos I was at that point of just, I’m not doing this anymore] and I think that was the point when I’d bonded with *child* and like, realised that I had a poorly child and he was my son, as opposed to just, this baby that I looked after | [laughs] so, errr, yeah... | P: How many months old was he then? | P: He was about three months | P: Three months? | P: I still hadn’t really held him, you know, he was, I think he was getting better at that point, yes, I think it was getting to the point that I was holding him, and I was... | P: He was becoming your son | P: He was becoming my son. And then I was like, oh shit! You know [laughs] this is actually really bad! Where, up to that point, I was just, I was sort of being the dutiful parent, doing all the right...
Appendix 17
Development of emergent and superordinate themes

<table>
<thead>
<tr>
<th>Page:Line</th>
<th>Original Transcript</th>
<th>Exploratory Comments</th>
<th>Emergent Themes</th>
<th>Superordinate Theme</th>
</tr>
</thead>
<tbody>
<tr>
<td>01:33</td>
<td>So I was looking after this child as you should, but I wasn’t upset, to the point that I should be</td>
<td>Doing the dutiful motherly thing, but more on a practical than an emotional level, so she was protecting her emotional wellbeing</td>
<td>manage emotional impact</td>
<td>control</td>
</tr>
<tr>
<td>02:01</td>
<td>I remember crying, and the midwife wrote on my notes “query post natal depression, mum upset”</td>
<td>Upset at being labelled, pathologised</td>
<td>manage emotional impact</td>
<td>control</td>
</tr>
<tr>
<td>2:03-2:04</td>
<td>Well, no, it’s just a bit emotional, I’m like 6 hours out of labour and you’ve got a bit of a query!</td>
<td>Defending her understandable emotional state, what do you expect?</td>
<td>manage emotional impact</td>
<td>control</td>
</tr>
<tr>
<td>2:04-2:05</td>
<td>But then after that I was fine, absolutely fine</td>
<td>Managing with the uncertainty in the hospital</td>
<td>manage emotional impact</td>
<td>control</td>
</tr>
<tr>
<td>2:05-2:06</td>
<td><em>child</em> erm, went into the ward, [WARD], and then he collapsed again the next day, so we ended up on ICU, and they decided to do surgery, but they couldn’t get a bed, and then he collapsed again, so they did surgery quicker than they’d planned</td>
<td>Rollercoaster of procedures, getting worse, getting better, systemic problems</td>
<td>management of condition</td>
<td>Acceptance</td>
</tr>
<tr>
<td>2:06-2:08</td>
<td>it was like, well yeah this is, this is what you do, you sit there with your baby and you do it, it was fine</td>
<td>Felt very routine, procedural, the new normal, what’s the point in questioning it?</td>
<td>management of condition</td>
<td>Acceptance</td>
</tr>
<tr>
<td>2:09-2:10</td>
<td>weaned the drugs wrong and things and he collapsed again</td>
<td>Even with the best care, things still can go awry</td>
<td>management of condition</td>
<td>Acceptance</td>
</tr>
<tr>
<td>02:12</td>
<td>So it all went on for like six months</td>
<td>This way of life became the new normal</td>
<td>integrate condition into lives</td>
<td>striving for normality</td>
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<tr>
<td>02:14</td>
<td>we were doing alternate nights</td>
<td>Working as a parent team</td>
<td>management of condition</td>
<td>control</td>
</tr>
<tr>
<td>Time</td>
<td>Event Description</td>
<td>Relevant Concepts</td>
<td></td>
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<td>02:15</td>
<td>and I just went home. I was like “I don’t want this anymore, I don’t want anything to do with my son, I don’t want anything to do with my husband!” [laughs] And I just went, I was like, that’s it!</td>
<td>Reached breaking point, couldn’t go on anymore, that was her limit</td>
<td>manage emotional impact</td>
<td>control</td>
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<tr>
<td>2:16-2:18</td>
<td>we hadn’t had any support at this point really, nobody really, you know, even the cardiac liaison nurses hadn’t become involved and we didn’t know anybody to help us</td>
<td>She had been doing it all alone, just parents struggling through, doing as best they could, unaware of what was available</td>
<td>need for support</td>
<td>Acceptance</td>
</tr>
<tr>
<td>2:20-2:21</td>
<td>Cos I was at that point of just, I’m not doing this anymore</td>
<td>Ready to give up, it’s too much to expect of a person</td>
<td>need for support</td>
<td>Acceptance</td>
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<tr>
<td>2:27-2:28</td>
<td>and I think that was the point when I’d bonded with <em>child</em> and like, realised that I had a poorly child and he was my son, as opposed to just, this baby that I looked after!</td>
<td>Moved from a detached caregiver to a mother, reality hitting and evoking care response</td>
<td>Parent responsibility</td>
<td>control</td>
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<td>2:28-2:29</td>
<td>I still hadn’t really held him</td>
<td>Delayed bonding, can’t bond with child until it feels safe to do so</td>
<td>Parent responsibility</td>
<td>control</td>
</tr>
<tr>
<td>02:34</td>
<td>was becoming my son. And then I was like, oh shit!</td>
<td>Reality setting in of what she could potentially lose, he wasn’t just a child anymore he was her child</td>
<td>Parent responsibility</td>
<td>control</td>
</tr>
<tr>
<td>02:37</td>
<td>this is actually really bad!</td>
<td>Realisation of what is at stake</td>
<td>diagnosis</td>
<td>Acceptance</td>
</tr>
<tr>
<td>2:37-2:38</td>
<td>, up to that point, I was just, I was sort of being the dutiful parent, doing all the right things</td>
<td>She had felt more removed, doing what was expected practically but not letting herself become emotionally involved</td>
<td>Parent responsibility</td>
<td>control</td>
</tr>
<tr>
<td>2:38-3:01</td>
<td><em>father</em> was the one who struggled, cos he, he did bond, and did, you know, immediately emotionally invest himself in this child, and worry about this child</td>
<td>Different response between parents, father became emotionally invested while mother did not</td>
<td>manage emotional impact</td>
<td>control</td>
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</tbody>
</table>