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Doctoral Thesis

# Parenting a child with congenital heart disease: Experiences of diagnosis, identity and parental role

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## Thesis Abstract

The thesis comprises a systematic literature review, an empirical research paper and a critical reflection on the research. The aim was to qualitatively explore parental experiences of having a child with congenital heart disease (CHD), specifically of receiving the diagnosis and of parental roles and identity.

The systematic literature review utilised thematic synthesis to synthesise qualitative findings regarding parents' psychological experiences of receiving their child's CHD diagnosis. Twenty-four papers were included in the review and four themes were identified: 1) unpreparedness for the diagnosis, 2) the overwhelming reality of CHD, 3) mourning multiple losses, and 4) redefining hopes to reach acceptance of CHD. There were individual differences in emotional experiences, but findings highlight the need for compassionate support from professionals throughout.

The research paper explored mothers' experiences of parental role and identity when parenting a child with single ventricle CHD (SVCHD). Interpretative Phenomenological Analysis (IPA) was used to analyse data from interviews with eight mothers. Four themes were identified: 1) being a "heart mum", 2) managing competing roles: "you have to wear lots of different hats all at the same time", 3) loss and regaining of identity, and 4) relinquishing control and the need to let go. Parenting a child with SVCHD presented significant challenges to mothers' parental role and identity, which they managed in various ways. Implications for clinical practice and future research are discussed.

The critical appraisal summarises the research and provides reflections on the methodological and ethical considerations, limitations and implications of the research. The appraisal highlights the importance of reflexivity, and personal reflections on the research process are considered throughout.

## Declaration

This thesis presents research undertaken for the Doctorate in Clinical Psychology at the Division of Health Research at Lancaster University from September 2019 to May 2022. The work presented here is my own, except where due reference is made. The work has not been submitted for any other academic award elsewhere.

Name: Shannon Dandy

Signed:

**Acknowledgements**

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## Contents

**Section One: Literature Review**


| | | |
|---|---|---|
| Title Page | | 1-1 |
| Abstract | | 1-2 |
| Introduction | | 1-3 |
| Method | | 1-6 |
| Results | | 1-9 |
| Discussion | | 1-20 |
| Conclusion | | 1-29 |
| References | | 1-30 |
| *Tables and Figures* | | |
| Table 1.1 | Search terms used for the systematic search | 1-44 |
| Figure 1.1 | PRISMA flow diagram depicting the study selection process | 1-45 |
| Table 1.2 | Characteristics of included studies | 1-46 |
| Figure 1.2 | Conceptual model of themes | 1-54 |
| *Appendices* | | |
| Appendix 1-A | Author guidelines for British Journal of Health Psychology | 1-55 |
| Appendix 1-B | Statements of contribution (as per journal guidelines) | 1-65 |
| Appendix 1-C | Example search strategy | 1-66 |
| Appendix 1-D | Scores on the quality assessment measure | 1-67 |
| Appendix 1-E | Example of transformation of line-by-line coding into descriptive and analytical themes | 1-69 |
| Appendix 1-F | Development of analytical themes | 1-71 |
| Appendix 1-G | Contribution of included papers to final themes | 1-72 |

**Section Two: Research Paper**


| | | |
|---|---|---|
| Title Page | | 2-1 |
| Abstract | | 2-2 |
| Introduction | | 2-3 |
| Method | | 2-6 |
| Results | | 2-10 |
| Discussion | | 2-19 |
| Conclusion | | 2-28 |
| References | | 2-30 |
| *Tables and Figures* | | |
| Table 2.1 | Participant demographic data | 2-41 |
| Figure 2.1 | Conceptual model of themes | 2-42 |
| Figure 2.2 | Participants' experiences of parental role and identity over time | 2-43 |
| *Appendices* | | |
| Appendix 2-A | Statements of contribution (as per journal guidelines) | 2-44 |
| Appendix 2-B | Murray and Wilde (2020) IPA methodology | 2-45 |
| Appendix 2-C | Example of coding | 2-46 |
| Appendix 2-D | Audit trail example: 'Emma' | 2-47 |
| Appendix 2-E | Contribution of each participant to the final themes | 2-49 |


**Section Three: Critical Appraisal**


| | | |
|---|---|---|
| Title Page | | 3-1 |
| Selecting the research topic | | 3-2 |
| Section 1: Systematic literature review | | 3-3 |
| | Summary of findings | 3-3 |

Methodological considerations 3-3

Reflections on the findings 3-5

Section 2: Empirical paper 3-6

Summary of findings 3-6

Methodological considerations 3-6

Ethical considerations: Managing distress 3-9

Data analysis 3-10

Reflexivity 3-11

Reflections on the findings 3-12

Personal reflections on the research process 3-13

Dissemination plan 3-14

Conclusion 3-15

References 3-16

**Section Four: Ethics Section**

Title Page 4-1

FHMREC Application Form 4-2

*Appendices*

Research Protocol 4-14

References 4-36

Appendix 4-A Participant information sheet 4-45

Appendix 4-B Social media advertisement 4-49

Appendix 4-C Covering email to charities 4-50

Appendix 4-D Text to be included in email advertisements 4-51

Appendix 4-E Expression of interest form 4-52

| | | |
|---|---|---|
| Appendix 4-F | Example email response to individuals not eligible to participate | 4-53 |
| Appendix 4-G | Microsoft Teams guidance | 4-54 |
| Appendix 4-H | Example email response to participants who wish to be interviewed as a couple | 4-58 |
| Appendix 4-I | Participant consent form | 4-59 |
| Appendix 4-J | Debriefing form | 4-61 |
| Appendix 4-K | Example semi-structured interview topic guide | 4-63 |
| FHMREC ethical approval letter | | 4-67 |
| FHMREC amendment correspondence | | 4-68 |

**Section 1: Systematic Literature Review**

# Parents' experiences of receiving their child's diagnosis of congenital heart disease: A qualitative systematic review

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**Abstract**

**Purpose:** This systematic review aimed to synthesise qualitative research regarding parents' psychological experiences following their child's diagnosis of congenital heart disease (CHD).

**Methods:** A systematic search of six electronic databases (CINAHL, Embase, MEDLINE, PsycINFO, PubMed and Web of Science) was completed, inclusive of all years up to May 2022. Sixty-seven full texts were screened against eligibility criteria. The included articles were synthesised using thematic synthesis and appraised using the Critical Appraisal Skills Programme Qualitative Checklist.

**Results:** Twenty-four articles were included in the review and four main themes emerged. Theme one, 'unpreparedness for the diagnosis' concerned parents' shock, guilt and anger regarding the diagnosis. Theme two, 'the overwhelming reality of CHD' described fear about decision-making and the child's prognosis, and the influence of professionals on parents' wellbeing. Theme three, 'mourning multiple losses', detailed parents' sadness at losing their envisioned pregnancy, birth and parenthood experiences. Theme four, 'redefining hopes to reach an acceptance of CHD' described parents' adjustment to the diagnosis.

**Conclusions:** Receiving a child's CHD diagnosis is a uniquely challenging situation for parents. The findings provided insight into the emotions parents experienced and how they adjusted to the diagnosis. As parents' experiences were significantly influenced by their interactions with professionals, clinicians should offer compassion, validation and clear information throughout the diagnosis process.

***Key words: Thematic synthesis; congenital heart disease; diagnosis; parent; psychological; emotional wellbeing.***

# Parents' experiences of receiving their child's diagnosis of congenital heart disease: A qualitative systematic review

Congenital anomalies are structural or functional defects present at birth, affecting 2-3% of babies in Europe (Morris et al., 2018). The diagnosis of a child's congenital anomaly can be a particularly stressful event (Wool, 2011) and usually occurs during the perinatal period: from conception to one year following childbirth. This is a time when parents are already at increased risk of experiencing the onset or relapse of mental health difficulties (Fairbrother et al., 2017; Howard & Khalifeh, 2020; Lee et al., 2007). Consequently, in the UK, where perinatal mental health has been prioritised, clinical psychologists are being integrated into maternity services to offer psychological support (British Psychological Society, 2016; National Institute for Health and Care Excellence, 2014). Similar practice is recommended globally: Australian guidance, for example, specifies regular assessment of perinatal psychological wellbeing and timely access to interventions (Austin et al., 2017).

Systematic reviews of literature show that perinatal psychological distress is more prevalent among parents experiencing 'high risk' pregnancies or births (Abrar et al., 2020; Biaggi et al., 2015), especially when there is concern about the child's health, including a diagnosis of a congenital anomaly (Wool, 2011). These parents face additional stressors, for instance, a review of 22 studies by Blakeley et al. (2019) documented the emotional struggle parents experience when deciding whether to continue or terminate the pregnancy following prenatal diagnosis of a life-limiting condition. Another review of 28 studies by Lou et al. (2017) concluded that severe prenatal diagnosis represents multiple, complex losses for parents, including that of a normal pregnancy, healthy child and envisioned future. It is, therefore, unsurprising that these parents are at increased risk of experiencing anxiety (Bekkhus et al., 2020), depression (Asplin et al., 2015) and post-traumatic stress (Bevilacqua et al., 2021; Cole et al., 2016), particularly at the time of diagnosis (Kaasen et al., 2017).

In qualitative research, parents describe various emotions following the diagnosis of a congenital anomaly, including shock, grief, guilt, anger and uncertainty (Carlsson et al., 2017; Hammond et al., 2020; Irani et al., 2019; Marokakis et al., 2017; Nelson Goff et al., 2013). In a review of 28 studies, Johnson et al. (2020) proposed a five phase model to represent parents' responses to discovering a foetal abnormality. The phases included expectations of the ultrasound, discovering the abnormality, intense shock, uncertainty and decision-making, and development of adaptive strategies to adjust to the diagnosis. However, there was little detail about what adjustment to the diagnosis involved. An earlier study by Lalor et al. (2009) presented a theory of 'recasting hope' to explain how mothers coped with foetal anomaly diagnoses. This process involved 'gaining meaning' by gathering information and making decisions, and 'rebuilding' by adapting previously held beliefs about pregnancy and their future. For those who terminated their pregnancy or whose child died, 'rebuilding' also involved grieving for their loss. The authors suggest that the 'rebuilding' phase could continue for several years following diagnosis of the congenital anomaly.

The most common congenital anomaly is congenital heart disease (CHD), which is a defect in the structure of the heart and has a global incidence of approximately 1% (Liu et al., 2019). There are several types of CHD, ranging from mild to severe, so exact treatment and prognosis vary. However, most children require at least one surgery and ongoing medical care (Hoffman et al., 2004). Although survival rates have improved, the experience of receiving a child's diagnosis is often distressing for parents, who face challenges such as understanding the condition, making medical decisions, and the possibility of their child not surviving (Hilton-Kamm et al., 2014). Therefore, there are significant psychological implications to consider for parents at the time of diagnosis, including how healthcare professionals support their understanding of information (Carlsson et al., 2015; 2016; Reid & Gaskin, 2018) and decision-making (Hoehn et al., 2004).

In a systematic review of 94 studies, Wei et al. (2015) highlighted diagnosis as particularly stressful for parents of children with CHD. Indeed, at the time of diagnosis, these parents have been found to score significantly higher on measures of stress, post-traumatic stress, anxiety and depression compared to clinical norms (Bevilacqua et al., 2013) and parents of healthy children (Brosig et al., 2007; Rychik et al., 2013). Some research suggests the timing of diagnosis is influential; for example, postnatal diagnosis has been associated with more anxiety and stress compared to prenatal diagnosis (Pinto et al., 2016). However, studies with larger samples have reported the opposite (Bratt et al., 2019) or no differences at all (Brosig et al., 2007). Crucially, these studies could not imply causation or, due to their quantitative nature, consider parental experiences in detail, such as how they respond, cope, and adjust over time. Thus, Wei et al. (2015) emphasised a need for further qualitative research.

In the past decade, many studies have qualitatively explored parents' experiences of having a child with CHD. Furthermore, systematic reviews have synthesised qualitative findings regarding parents' psychosocial coping (Lumsden et al., 2019), fathers' perspectives (Lin et al., 2021) and the wider familial impact (Jackson et al., 2015), as well as parents' experiences of specific events, such as surgery (de Man et al., 2020; McMahon et al., 2020). Tacy et al. (2022) described current practices of prenatal counselling by medical professionals at diagnosis, highlighting the importance of clear information and compassion. However, despite diagnosis being crucial in parents' experiences of their child's CHD, there has been no review integrating relevant qualitative research findings. A review of this nature is vital to understanding parental experiences and support needs, especially given recommendations for accessible psychological support at the time of diagnosis (Gramszlo et al., 2020; NHS England, 2016). Therefore, this review and meta-synthesis aimed to systematically review and synthesise qualitative evidence to answer the following research

question: what were parents' psychological experiences when they received their child's CHD diagnosis?

## Method

The review was completed in accordance with Preferred Reporting Items for Systematic Reviews and Meta-Analysis (PRISMA) guidance (Moher et al., 2009) and was registered on PROSPERO (Ref: CRD42021264117).

### Search Strategy

The search strategy was developed in consultation with the research team and specialist subject librarians. Initial scoping searches of keywords related to the research question (e.g., parent, CHD, diagnosis, and qualitative methods) were completed to inform the final search strategy.

A systematic search of six databases (CINAHL, Embase, MEDLINE, PubMed, PsychINFO and Web of Science) was initially completed in January 2022 and updated in May 2022, inclusive of all years up to 17 May 2022. No new papers were identified by the second search. The SPIDER search tool (Cooke et al., 2012) was used to select and organise search terms (see Table 1.1). The terms were combined using Boolean operators 'OR' within each concept and 'AND' between concepts. Despite being central to the research question, the word "diagnosis" was not included in search terms because scoping searches found this excluded articles exploring wider parent experiences, of which diagnosis was only part. Medical Subject Headings (MeSH) and exploded terms were used when possible. Searches were limited to title and abstract. To improve recall of papers, a near field search was included (i.e., heart N5 (disease OR defect)), because qualitative papers may not use fixed terminology (see appendix 1-C for a full search strategy example). Finally, reference lists of

included papers were checked and the 'cited by' function on Google Scholar was used to identify any additional papers.

[INSERT TABLE 1.1 HERE]

**Eligibility Criteria**

To be included in the review, studies had to meet the following inclusion criteria: 1) had a sample consisting of parents of children with a diagnosis of CHD, 2) included sufficient findings[1] related to parents' psychological experiences when receiving their child's CHD diagnosis, 3) utilised qualitative methods for primary data collection (mixed methods studies were considered if a qualitative methodology was described and sufficient qualitative data were presented), 4) used a recognised, inductive method of qualitative analysis, 5) explored first person accounts, evidenced by original data excerpts, 6) were published in a peer reviewed journal and 7) were available in English or German (languages the authors were fluent in). Studies were excluded if they 1) focused on other physical health conditions as well as CHD or 2) were a literature review.

**Management of the Selection Process**

The selection process is illustrated in Figure 1.1. Search results were collated using EndNote referencing software. The first author screened all titles and abstracts against eligibility criteria and repeated this process with full texts. To ensure reliability in the selection process, two independent reviewers (a trainee clinical psychologist and an assistant psychologist with master's level research training) reviewed 10% of titles and abstracts (n=146) and 25% of full texts (n=6). The agreement between researchers was 98.63% (n=144) and 83.33% (n=5), respectively. Disagreements were resolved through discussion.

[1]Sufficient findings was defined as at least one quote and one author interpretation specifically referencing parents' emotional experiences at the time of, or in relation to, receiving their child's CHD diagnosis.

[INSERT FIGURE 1.1 HERE]

**Quality Appraisal**

The methodological quality of included studies was assessed using the Critical Appraisal Skills Programme Qualitative Checklist (CASP) (2018). The checklist comprises two screening questions, followed by eight items considering various aspects of study quality, including research design, data collection and ethical issues. A three-point scoring system was adopted for each item, as per Duggleby et al. (2010): a score of 1 represented weak evidence, 2 represented moderate evidence, and 3 represented strong evidence. Methodological quality of all studies was assessed by the first author and an independent researcher (a trainee clinical psychologist), to ensure accuracy.

The maximum possible score was 24, and scores for included articles ranged from 14 to 24 (mean=19.25, SD=2.37) (see Appendix 1-D). The agreement on ratings between the first author and independent reviewer was 100%. A particular weakness of the included studies was consideration of the researchers' role and potential bias, which only four articles explicitly referred to. However, this may reflect reporting conventions in peer reviewed journals (e.g., word limits), rather than the quality of the research process itself (Majid & Vanstone, 2018).

No papers were excluded based on the quality assessment, in the context of ongoing debates and lack of consensus regarding what constitutes quality in qualitative research and how this should be measured (Garside, 2014). Instead, the review sought to provide a thorough overview of the current research landscape, with quality appraisal used to critically consider each study and its contribution.

### Data Extraction and Synthesis

Key characteristics from eligible studies were extracted and tabulated, including authors, year, country, title, research aims, sample size and characteristics (gender, age, ethnicity, severity of child's diagnosis), data collection method, analysis method and results.

Thematic synthesis, as described by Thomas and Harden (2008), was used to synthesise data. The approach was chosen following consideration of guidance for qualitative evidence synthesis, because thematic synthesis utilises methods from meta-ethnography and grounded theory to enable synthesis of both descriptively 'thin' and 'rich' data, whilst still allowing for interpretation (Booth et al., 2016; Flemming & Noyes, 2021).

Data were analysed inductively using NVivo software. In accordance with Thomas and Harden (2008), 'data' were considered to include all text under 'results' or 'findings' headings. Firstly, the first author repeatedly read each study and completed line-by-line coding. As the same process was completed for each included study, reciprocal translation was possible at this stage. Secondly, codes were grouped into descriptive themes that represented similar concepts. Finally, the first author compared and interpreted the descriptive themes to develop analytical themes, which were discussed and amended alongside the research team. Evidence of how descriptive and analytical themes were derived from initial codes is documented in Appendices 1-E and 1-F.

## Results

### Study Characteristics

In total, 24 papers published between 1995 and 2021 were included in the synthesis. Sample sizes ranged from 8 to 34 participants (mean=15.71, SD=8.44), representing 377 parents and comprising 226 mothers and 141 fathers (one study included 10 parents, but did

not specify mothers and fathers). The key characteristics and themes of each paper are outlined in Table 1.2.

Nineteen studies collected data using semi-structured interviews, one used focus groups (Leuthner et al., 2003) and another used open questions in a questionnaire (Williams et al., 2019). Two studies used mixed methods (McKechnie et al., 2016; Williams et al., 2019), but only qualitative data were extracted. Included papers used inductive analysis, including thematic analysis (n=8), content analysis (n=5), thematic content analysis (n=3), grounded theory (n=3), descriptive phenomenology (n=2) and interpretative phenomenological analysis (n=1). Two studies used directed content analysis and, although this is a deductive method, the authors state that the analysis was not limited to predefined categories (McKechnie & Pridham, 2012; McKechnie et al., 2016). Following discussion with a research team member with expertise in qualitative research (CM), it was agreed to include the studies in the review.

[INSERT TABLE 1.2 HERE]

Four main themes reflecting parents' psychological experiences regarding their child's CHD diagnosis, each with subthemes, were identified: 1) unpreparedness for the diagnosis, 2) the overwhelming reality of CHD, 3) mourning multiple losses and 4) redefining hopes to reach acceptance of CHD. A narrative summary of each theme is provided below, and Appendix 1-G illustrates how each study contributed to these. Figure 1.2 presents a conceptual model of themes, which appear to represent phases of emotion that evolve over time. Whilst shock and unpreparedness for the diagnosis appeared to occur immediately, parents' experiences of the other phases were not necessarily linear and could

continue for weeks, months or years following the diagnosis. Furthermore, some parents experienced all of the emotions described, whilst others only experienced some.

[INSERT FIGURE 1.2 HERE]

**Theme 1: Unpreparedness for the diagnosis**

Parents' initial unpreparedness for their child's CHD diagnosis was detailed in 20 papers, alongside emotions of shock, guilt and anger. One parent encapsulated the experience: "It [the diagnosis] was the most devastating thing I have ever heard in my life" (Wei et al., 2016, p.156). Several studies also reported short-term avoidant coping strategies.

***Subtheme 1.1: Shock***

Powerful shock and feelings of dismay at diagnosis related to parents' assumptions that nothing would be wrong with their child: "It's a big shock when it happens because you think you're going to have a perfectly healthy baby, when they tell you it's a serious condition" (Messias et al., 1995, p.370). Consequently, studies described the experience as 'surreal', as one parent likened it to: "a scene in a TV drama" (Hwang & Chae, 2020, p.110). Another conveyed the trepidation and lack of control parents felt: "…after only hearing about such things on TV – I, or people like us, thought that a heart transplant would be needed or something like that. I felt as if I was falling off a cliff" (Kim & Cha, 2017, p.441). The unexpected nature of the diagnosis heightened emotions of powerlessness and devastation, leading some parents to experience visceral symptoms: "The first time I was told about my child's disease I was unconscious for an hour and a half" (Nayeri et al., 2021, p.4).

Throughout, postnatal diagnosis seemed especially shocking. A 'normal' pregnancy may have fostered a sense of security that postnatal diagnosis disrupted, leading to confusion

and increased distress. Indeed, Thomi et al. (2019, p.4) note these parents had no time to process the diagnosis before treatment began: "Suddenly an entire team of nurses, midwives and physicians were treating their new-born".

### *Subtheme 1.2: Blame towards self and others*

Parents immediately searched for causes of their child's CHD. Mothers especially experienced intense guilt, because they felt responsible for carrying the child: "I was blaming myself for a lot of things… What did I do wrong for him to be this way? Like I just feel like, what did I do that my son's so sick?" (Sood et al., 2018, p.637). There were some cultural variations, as Im et al. (2018, p.470) found that inability to meet Korean cultural expectations to product a 'perfect' child heightened maternal guilt and created additional shame: "She [participant's sister] said: 'you'll be able to be pregnant easily next time, so why do you want to deliver an imperfect baby?'".

Despite blaming themselves, mothers felt resentful that they had done everything 'right': "I was annoyed, cos I didn't smoke, I didn't drink. I didn't do anything… I just don't understand why I got it" (Lumsden et al., 2020, p.4). Conversely, fathers appeared to experience more anger towards the world, implying recognition that the diagnosis was out of their control: "For myself, I think I felt a lot of anger. 'Why us?'… 'Why does my wife have to go through this?' I had a lot of anger at God" (Leuthner et al., 2003, p.124). The intensity of their anger also appeared to result from feeling their partner's pain as well as their own.

### *Subtheme 1.3: Avoidance*

Avoidance was a common theme in parents' initial response to the diagnosis. Some experienced a sense of incredulity or feelings of numbness: "My husband said baby is healthy and don't need to seek treatment. The doctors are wrong" (Nayeri et al., 2021, p.36). This

denial of CHD or its severity may have protected against the emotional impact of diagnosis. Similarly, some studies suggested parents withdrew socially to avoid sharing the diagnosis and associated emotions with others (Leuthner et al., 2003; Nayeri et al., 2021). However, McKechnie and Pridham (2012, p.1699) postulate that, for many parents, this was adaptive and allowed space to confront their emotions of sadness, fear anxiety and confusion: "Parents described their efforts to sort out their emotions by first talking with their significant other and then expanding the talking to family members, other close contacts, and support groups".

**Theme 2: The overwhelming reality of CHD**

The second theme describes intense emotions once parents realised the severity of CHD, detailed in 19 papers. These emotions included feeling overwhelmed by information, uncertainty, and fear regarding their child's prognosis. Parents coped in various ways, but their experiences seemed especially affected by interactions with professionals.

***Subtheme 2.1: Overwhelming information and decision-making***

Whilst trying to manage initial shock, parents had to navigate information about CHD and treatment decisions. This experience was often overwhelming and frightening: "…it's all one big blur. It's just so much to take in and like, understand" (Bertaud et al., 2020, p.1227). Parents, particularly those receiving a postnatal diagnosis, felt unprepared for this and under pressure to 'do the right thing': "You're in shock… you had a Caesarean, you haven't got time to wrap your head around stuff, and you're told all this information and told you need to make decisions quickly" (Cantwell-Bartl & Tibballs, 2015, p.1069). Nonetheless, across studies, parents valued decisions being theirs to make because it made them feel respected and in control: "It was always left to me to decide what I wanted to do rather than being pushed into anything" (Bertaud et al., 2020, p.1227).

However, those who did not understand CHD expressed self-criticism, shame and fear about decision-making: "The doctor explained everything, but I couldn't understand a word he was saying because I didn't understand medical words. I thought I would look stupid if I asked" (Cantwell-Bartl & Tibballs, 2015, p.1067). Similar concerns were expressed across studies from different regions, including the USA and Iran: "We were upset that we didn't understand my son's illness" (Nayeri et al., 2021, p.36), suggesting commonalities in experience despite differing cultures and health systems.

### *Subtheme 2.2: Uncertainty and fear*

Whilst parents received information about CHD, uncertainty regarding the prognosis remained, so anxiety was prevalent: "The [diagnosis] was scary. It was overwhelming, lots of uncertainty, his diagnosis came before he was born so we still had about five months of anticipation" (Neubauer et al., 2020, p.1674). This led to intense fear and rumination about their child's survival: "My stress levels were high, and I was nervous as to how this was all going to play out for [child] when he was born" (Williams et al., 2019, p.930). Another parent stated: "We didn't know whether she would make it or not" (Messias et al., 1995, p.372).

Parents felt hopeless at being unable to help their child and three papers that included both mothers and fathers reported that hopelessness appeared more prominent for mothers (Leuthner et al., 2003; Messias et al., 1995; Sood et al., 2018). One parent recalled: "I had the feeling that this child will not survive. We'll just lose it" (Thomi et al., 2019, p.4). Even those with a postnatal diagnosis, who had less time to ruminate on CHD, expressed similar fears. Rempel et al (2013, p.622) detailed that one mother: "described feeling 'more scared than anything' and devastated that her baby was 'dying of course'". Pessimism was mainly reported in relation to severe CHD, when death is more likely, so may have been a way to

manage expectations or distress. Indeed, Lee and Ahn (2020, p.7135) found mothers who felt unable to manage their hopelessness could become suicidal: "It was too hard for me. At that time, I even thought about dying with my kid who might die anyway".

### *Subtheme 2.3: Coping and the role of professionals*

Parents coped with fear and uncertainty by hoping that their child would survive: "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" (McKechnie & Pridham, 2012, p.1700). Some also sought control by, for example, gathering information: "That week I just was closed down, I just wanted to search and make sure I was doing the right thing" (Bertaud et al., 2020, p.1227). For fathers, being in control was viewed as important in carrying out gendered social roles, such as suppressing their emotions to support their partner: "I didn't want my girlfriend to see me crying, since [I'm supposed] to try to be strong for her" (Clark & Miles, 1999, p.11). Thus, their experiences of the diagnosis may go unrecognised by family or professionals.

Parents' experiences of, and coping with, the diagnosis were materially affected by interactions with professionals. Clear information and treatment plans, with opportunities for repetition, were valued because they allowed parents to feel reassured, thereby reducing stress: "They went through it over and over again, they did diagrams, they gave us information for charities… If they hadn't have done all that I don't think I would have coped" (Bertaud et al., 2020, p.1227). There was also a sense of relief and hopefulness: "Her diagnosis was terrifying but the steps and treatment plan in place did make [me] feel positive about her future" (Williams et al., 2019, p.930).

Parents especially valued compassion at a time when they felt extremely vulnerable: "We were encouraged to ask questions, we were informed very well by different people, by

nurses and physicians. We were involved and taken seriously… not only our baby but also we were important" (Thomi et al., 2019, p.6). Indeed, parents who found such qualities lacking in their interactions with professionals felt angry, dismissed and increasingly anxious: "We were kind of squeezed in where, I would say, it felt rushed. We didn't have the time or the knowledge to ask everything we wanted to" (McKechnie et al., 2016, p.84).

**Theme 3: Mourning multiple losses**

The third theme, regarding multiple losses brought by the CHD diagnosis, was detailed in 16 papers. Parents described that they lost 'normal' experiences of pregnancy and birth, which became overshadowed by aforementioned uncertainty and fear. They also mourned the loss of a healthy child and their envisioned future; for example, feeling the diagnosis would impede fulfilment of previously held expectations of parenthood.

***Subtheme 3.1: Loss of 'normal' pregnancy experiences***

Parents who received a prenatal diagnosis recalled losing a 'normal' pregnancy. Joy about the pregnancy was difficult to reconcile with the seriousness of CHD, so typically exciting pregnancy experiences became tainted by sorrow and concern: "Every kick, every push, every movement, I don't know how I felt. I felt bad for myself, but worse for my wife, because she felt every single thing, every day" (Leuthner et al., 2003, p.125). Consequently, parents felt despair at being unable to revel in pregnancy rituals, despite these being longed for: "The diagnosis left them 'heartbroken' and shifted their attention away from joyful future plans like 'putting a nursery together'" (Harris et al., 2020, p.8).

Mothers expressed further sadness at losing typical birth experiences, which they seemed to ruminate on throughout pregnancy: "They're [nurses] gonna get to know your baby and all of the little idiosyncrasies that the baby's gonna have and I want that to be me"

(McKechnie et al., 2016, p.88). Mothers and fathers receiving a postnatal diagnosis felt a more sudden loss of their birth experience that created stress and fear: "It was so unrealistic, a horror scenario. Finally, the child is here and one minute later, they [the HCPs] again take her [baby] away" (Thomi et al., 2019, p.4). Conversely, fathers receiving a prenatal diagnosis appreciated opportunities to prepare for a different birth: "Time, to look up our doctors, doctors that would follow his condition… And how the [hospital] operates, to prepare for it. So that really helped" (McKechnie & Pridham, 2012, p.1702), suggesting they had a more practical focus.

***Subtheme 3.2: Loss of a healthy child and envisioned future***

Regardless of timing, the CHD diagnosis was often a devastating experience accompanied by emotions of grief and sorrow because parents lost their expected healthy child: "It was like mourning the pregnancy that I thought we would have… mourning the overall health of my baby boy" (Espinosa et al., 2021, p.5). Another parent described: "I went through these stages of, I don't know, it almost felt like stages of grief even though nobody had died but maybe it was my perception of the ideal child did die" (Woolf-King et al., 2018, p.2789). However, gender expectations to be 'strong' meant fathers often felt unable to express and process their grief, leading them to feel isolated: "I wept a lot while being seated at the back of the bus and walking through our apartment complex. It was hard to endure my sadness" (Hwang & Chae, 2020, p.110). Carlsson and Mattsson (2018, p.30) interviewed parents who terminated the pregnancy following diagnosis, and suggested their loss was more concrete and compounded by guilt: "The pregnancy termination involved considerable emotional stress and the loss of a wanted child, likened by respondents to an execution".

Losing the health of their child also disrupted parents' view of themselves as parents: "We became aware that, yes, we actually have a very sick child. Even if it [child] has good

chances, it is not a healthy heart, but a repaired heart" (Thomi et al., 2019, p.4). This disruption created pessimism and anxiety about the future: "I didn't think I'd be able to do anything, that I'd have to walk on eggshells, and that I wouldn't enjoy him" (McKechnie & Pridham, 2012, 1701). In response, parents hoped that their child might still have a good quality of life and fulfil some expectations: "That generally her life would be normal, but that she may be limited in the physical activities/exercise she can engage in" (Williams et al., 2019, p.929).

**Theme 4: Redefining hopes to reach acceptance of CHD**

The final theme reflected parents' coping and adjustment to the diagnosis, identified in 14 papers. Some coping strategies applied to specific situations, as above, and others were utilised throughout. Nonetheless, these strategies appeared to support parents to adjust their expectations of pregnancy, their child and future. However, the strategies and time taken to adjust varied.

***Subtheme 4.1: Ongoing coping strategies***

Parents' coping depended upon individual responses to the diagnosis. However, most discussed support from others throughout: "Having good family support has been helpful. I'm very emotional in a crisis and my husband's a calm person. That's been helpful" (Espinosa et al., 2021, p.5). Parents often emphasised the value of peer support, which seemed to offer a unique, shared understanding: "I met many mothers and their children who were in the same boat, with the same pain and grief… We all comforted one another" (Lee & Ahn, 2020, p.7138). Harris et al. (2020, p.9) reported that some parents worried that peer support created "false hope" but was the only study to do so. Conversely, across studies, many parents used religion to understand and accept the diagnosis: "We said this child is

created by God. If we terminate, it is like interfering with God's will. What God has given us we have to accept" (Cantwell-Bartl & Tibballs, 2015, p.1070).

### *Subtheme 4.2: Adjusting expectations of pregnancy*

As parents with a prenatal diagnosis processed their emotions, they were able to "reframe the pregnancy as a personable and enjoyable experience" (McKechnie et al., 2016, p.86). This promoted engagement in pregnancy rituals (e.g., nursery decoration), that allowed parents to feel content and further facilitated adjustment. Although some were reluctant to accept CHD, doing so seemed to create capacity for hope: "There is no way to avoid the given fate. I guess I just have to accept it. If I go to the hospital with the child and go after it hard, I believe it could get better someday" (Lee & Ahn, 2020, p.7139).

There were also narratives of making the most of pregnancy: "You kept on going. You had to enjoy your pregnancy, 'cause if you didn't, you were just going to make things worse" (Rempel et al., 2013, p.622). Mothers sought connection with their child, which provided comfort and happiness: "…when she told me, I made the decision that every minute I have this child alive inside me is a moment to cherish." (Leuthner et al., 2003, p.125). Similarly, Im et al. (2018, p.471) found Korean mothers were influenced by *TaeKyo*, a traditional concept that suggests connection with the foetus supports its development. These mothers exercised emotional resilience to interact with their child, which facilitated acceptance of their CHD: "I felt happy when she was kicking my belly. I felt as if she was sending me a signal that 'Mommy, I'm here, and I will be OK."

### *Subtheme 4.3: Redefining hopes for the child*

Parents' adjustment to the diagnosis was evidenced as they redefined hopes for their child and parenthood. Some prepared to promote fulfilment for their child, despite CHD, as

they appeared to feel excited and thankful for their child's future: "We were already getting ready for what kind of things we could expose him to, piano lessons, or… Whatever we could throw his way to help him live it to the fullest" (Neubauer et al., 2020, p.1674). However, most discussed defining a 'new normal' and adapting their life to accommodate CHD: "Participants formed a new family identity incorporating the prenatal diagnosis" (Harris et al., 2020, p.8). A narrative emerged regarding parents' unconditional love for their child and, consequently, reappraising their priorities for parenthood. One mother discussed promoting bonding opportunities: "…Especially with the surgery and knowing that it's going to be open-heart. How can we bond? We'll figure out some way so that we can still breastfeed" (McKechnie & Pridham, 2012, p.1701). Similarly, fathers discussed adapting interests to share with their child: "[If] having a heart condition definitely limits his ability to participate, he could still just enjoy being a fan or learning about the sports" (Harris et al., 2020, p.8).

## Discussion

The systematic literature review aimed to synthesise qualitative findings regarding parents' psychological experiences when receiving their child's CHD diagnosis and is the first systematic review to do so. Four themes were identified to encapsulate parents' experiences. Parents were unprepared for the shock of the diagnosis and felt guilty and angry (theme 1). They were faced with overwhelming information, decision-making and uncertainty regarding their child's prognosis. These experiences were especially influenced by interactions with professionals (theme 2). The diagnosis and associated disruption led parents to mourn the loss of a 'normal' pregnancy or birth and of a healthy child (theme 3). However, parents used various coping strategies throughout, facilitating their adjustment to the diagnosis (theme 4).

The synthesis identified that parents experienced powerful emotions following their child's CHD diagnosis, including shock, guilt, anger, fear and uncertainty. This supports previous findings related to the diagnosis of other congenital anomalies (Carlsson et al., 2017; Hammond et al., 2020; Irani et al., 2019; Marokakis et al., 2017; Nelson Goff et al., 2013). Interestingly, the synthesis revealed a narrative that these emotions, and the seriousness of the CHD diagnosis, were difficult to reconcile with the joy parents had initially felt towards their pregnancy or new-born. Their subsequent distress could be explained by theories of cognitive dissonance, which suggest incompatible thoughts about an experience or phenomenon can create unpleasant psychological states (Harmon-Jones & Judson, 2019). Due to the uncertain prognosis, parents were unable to immediately resolve their simultaneous sorrow and happiness, so felt unable to enjoy typically exciting experiences (e.g., feeling the baby kick).

The diagnosis also seemed to represent multiple losses for parents, of a 'normal' pregnancy or birth, healthy child and envisioned future. Experiences highlighted throughout the synthesis, of anger, avoidance, sorrow and eventual acceptance of the diagnosis, are analogous to those described in Kübler-Ross and Kessler's (2005) grief cycle. This model details the 'natural' process of grief: denial, anger, depression, bargaining and acceptance, suggesting that the experience of receiving a child's CHD diagnosis might have similarities with that of a concrete loss following death. In their review of literature, Lou et al. (2017) similarly found that prenatal diagnosis of a lethal condition represented multiple complex losses for parents. Whilst CHD is a serious condition, severity and prognosis varies. The studies included in the synthesis represented various CHD types, suggesting that concepts of grief and multiple losses for parents also apply to diagnoses that are not lethal.

Although there were consistencies across parents' experiences, the synthesis identified some differences, particularly according to gender and timing of diagnosis. Across

studies, fathers were less likely to express their emotions due to gender norms and expectations to be 'strong' and care for their partner. This finding was consistent across cultures and reflects previous research regarding fathers' experiences of their child's CHD more generally (Gower et al., 2017; Lin et al., 2021).

With regards to timing, quantitative research had identified differences in levels of distress between parents who receive a prenatal or postnatal diagnosis (Bratt et al., 2019; Pinto et al., 2016). The current synthesis expands these findings to include qualitative detail about parents' differing experiences that was consistent across multiple studies. Those receiving a prenatal diagnosis described more intense rumination and uncertainty throughout pregnancy, creating anxiety and hopelessness. Conversely, those receiving a postnatal diagnosis experienced more intense shock and feelings of being overwhelmed, because no previous concerns had been identified and parents had to navigate multiple emotions alongside time-pressures to make medical decisions. Consequently, the synthesis illustrates that the diagnosis of a child's CHD may be universally distressing for parents, but the specific emotions involved and timepoints at which psychological wellbeing is affected varies between individuals.

Parents' experiences of, and preferences for, information following their child's CHD diagnosis have been previously discussed (Carlsson et al., 2015; Reid & Gaskin, 2018). The synthesis offered consistent findings that parents find information about their child's CHD overwhelming, yet they also feel an obligation to learn as much as possible. Furthermore, positive interactions with professionals appeared to be a key influencer of parents' psychological wellbeing because they valued validation and opportunities to ask questions or hear information repeated. Previous literature reviews have similarly shown that professionals' personable skills can facilitate parents' coping with their child's congenital anomaly diagnosis (Kratovil & Julion, 2017; Lou et al., 2017). However, the current

synthesis extends these findings: there were examples of unhelpful practice across studies, such as professionals lacking compassion or rushing information sharing. This illustrates that professionals can actively, albeit negatively, influence parents' psychological wellbeing.

Across the included studies, parents' use of coping strategies was discussed, including avoidance, hope, seeking control, and support from others. These strategies were similar to those described in a previous systematic review exploring psychosocial coping among parents of children with CHD (Lumsden et al., 2019), which included five papers from the current review. The findings may be conceptualised using Lazarus and Folkman's (1987) Transactional Model of Stress and Coping, which suggests that coping is determined by an individual's appraisal of the threat posed by a stressor and of their available options. The model proposes two types of coping: problem-focused coping seeks to address the problem, whilst emotion-focused coping prioritises managing the emotional response. Thus, in the current synthesis, parents may use emotion-focused coping strategies, such as denial, hope or avoidance to appraise their child's diagnosis as less threatening and protect them from the psychological implications of CHD. Other parents may appraise the diagnosis as highly threatening and use problem-focused coping strategies, such as seeking control or information, to try to address problems, such as decision-making. There may be wider psychological, social, practical or cultural variables that inform parents' appraisal of the diagnosis and subsequent coping that were not identified by the synthesis.

Nonetheless, parents' use of coping strategies appeared to facilitate overall adjustment to the CHD diagnosis. Lalor et al. (2009) proposed 'recasting hope' as a model of adjustment following prenatal diagnosis of a congenital anomaly. This model involved a phase of 'rebuilding' by seeking meaning in the diagnosis and reconstructing hopes for the future. The current review presents similar findings that parents adjusted their expectations and reappraised their hopes for the child and future in the context of CHD. However, the

synthesis revealed that this process of adjustment was facilitated by an overarching narrative of unconditional love for their child, which has not been previously reported.

Taken together, the themes identified in the current synthesis appear to evolve over time and may map onto Johnson et al.'s (2020) model of parents' responses following prenatal diagnosis of various foetal anomalies. The model involves five phases: 1) expectations of scans, 2) discovery of the anomaly, 3) shock, 4) decisions and planning, and 5) adaptation.

Two studies included in the review proposed similar models that the findings of the synthesis could map onto. Firstly, Im et al. (2018) suggested that mothers experienced four phases following their child's prenatal CHD diagnosis: 1) shock and pain, 2) worries and concerns, 3) recognition of the child as a living being, and 4) restructuring the pregnancy experience. However, this model did not consider the experiences of fathers or those with a postnatal diagnosis. Secondly, according to Neubauer et al. (2020), parents experienced six transitions from receiving their child's CHD diagnosis to their death: 1) learning the diagnosis prenatally, 2), learning the diagnosis postnatally, 3) new normal, 4) taking control, 5) learning death is likely, and 6) after death.

Whilst the shock, fear, decision-making and adjustment described in the aforementioned models are similar to the findings from the synthesis, the current findings expand on these models to include additional experiences of anger and uncertainty, as well as consideration of parents' coping at each phase following diagnosis. Furthermore, the findings from this review include a phase of mourning, as children with CHD require ongoing care and adaptation, which was not represented in other models. Finally, the themes from the current synthesis considered individual differences that previous models have not, such as between mothers and fathers, those prenatally compared to postnatally diagnosed, and across various types of CHD and cultural contexts.

**Clinical Implications**

Many parents reported powerful distress following their child's CHD diagnosis, but the extent to which they struggled or coped with this varied. Therefore, as recommended by Blakeley et al. (2019), a stepped care model of psychological support may be beneficial. All staff could offer compassion, validation, normalisation and basic psychoeducation. Their ability and confidence to do so could be enhanced by teaching or consultation from clinical psychologists. A smaller number of parents who experience greater or continued difficulties could then access individual support from a clinical psychologist, consistent with recommendations for CHD services (NHS England, 2016).

It is important to note the potential emotional impact of delivering diagnoses of congenital anomalies on health professionals themselves, who have been reported to suffer from burnout and compassion fatigue (Cohen et al., 2020; Tacy et al., 2022). This could explain the negative interactions with professionals that some parents report. Thus, staff who deliver a child's CHD diagnosis to parents, or offer prenatal counselling, may benefit from access to regular supervision or reflective practice facilitated by a clinical psychologist, to support their own wellbeing and self-awareness. Indeed, similar practice has been reported, and positively evaluated, throughout health services (Beavis et al., 2021; Davey et al., 2020).

Although many parents reported shared experiences, it should not be assumed that parents experience all of the emotions described in the synthesis in the same way, or at all. Indeed, variation between individuals was highlighted, so professionals should consider parents' individual needs and tailor support to these. For instance, differences were highlighted between parents who received a prenatal compared to postnatal diagnosis. Those receiving a prenatal diagnosis reported rumination on loss of their expectations of a healthy child, so may benefit from support with prenatal bonding. Conversely, those who received a

postnatal diagnosis reported more shock, so may benefit from support to manage and process information.

The synthesis also highlighted differences in fathers' experiences, compared to mothers', because they felt under pressure to remain 'strong' and suppress their emotional responses to the diagnosis to care for their partner. Similar findings have been identified in previous research regarding fathers' broader experiences of CHD (Lin et al., 2021). Professionals should, therefore, encourage fathers to share their emotions and experiences.

Parents used various coping strategies following the diagnosis and should be encouraged to draw upon their existing resources to facilitate adjustment. Peer support was highlighted throughout the synthesis as valuable due to the normalisation it provided. Use of peer support has been found to be beneficial for parents of children with a range of conditions (Bray et al., 2017; Lumsden et al., 2019; Shilling et al., 2013). Health services could, therefore, promote further access to peer support by facilitating groups for parents who receive a diagnosis of CHD for their child, either in person or online. Clinical psychologists could be involved in running these groups to offer psychoeducation, normalisation and validation regarding parents' psychological responses to the diagnosis, as these factors have been identified as key to therapeutic relationships and positive therapeutic outcomes (Wampold, 2015; Yuen et al., 2022). They could also facilitate discussions between parents to support the development of shared understanding, and identify any parents who may benefit from additional, individual support.

**Strengths and Limitations**

This was the first review to synthesise qualitative literature regarding parents' psychological experiences of their child's CHD diagnosis. Findings provided an essential summary of the emotional trajectory these parents may experience, which can inform support

offered by health services. Findings were consistent across studies, illustrating that the synthesis was not predicated on one population (e.g., Western) or on studies of lower quality.

However, a key limitation was the subjectivity inherent to qualitative literature synthesis, which relies on the author(s) to select, appraise and synthesise studies. For instance, criteria for studies to be published in a peer-reviewed journal may have omitted other findings containing alternative perspectives. Nonetheless, efforts were made to ensure rigour, as independent researchers supported the selection and quality appraisal processes. Although coding and identification of descriptive themes were completed by the first author, final themes were identified through collaborative discussion with the research team. Furthermore, Appendices 1E-1G illustrate how final themes were identified, providing evidence to support the credibility of the analysis.

A further limitation was the application of numerical criteria to the CASP to appraise the quality of included studies, because it was not designed for this purpose (Majid & Vanstone, 2018). However, the reliability of scores was enhanced by the independent reviewer and no studies were excluded on the basis of this scoring. Instead, it provided a clear indication of the quality of included studies that could inform theme development and the presentation of supporting data.

Finally, 19 of the included studies contained fathers' perspectives. This contrasts with the wider research area, which has predominantly focused on mothers (Wei et al., 2015). However, findings revealed that fathers often felt unable to express their emotions related to their child's CHD diagnosis, due to gender expectations to be 'strong', so the synthesis may not provide an accurate summary of their experiences.

**Future Research**

The systematic review provided insight into parents' qualitative experiences of receiving their child's CHD diagnosis. However, there has been no published review of the quantitative research in this area. A quantitative systematic review would likely represent a greater number of parents and offer findings that were not possible in this qualitative review, such as comparison with other groups (e.g., parents of healthy children). A review of this nature is essential to augment our understanding of parents' psychological needs at the time of diagnosis and how best to support them.

The synthesis highlighted the role of professionals in shaping parents' experiences of their child's CHD diagnosis, for example, through compassionate communication. Previous research has considered parents' informational needs at diagnosis (Carlsson et al., 2015) and Tacy et al. (2022) provided an overview of prenatal counselling practices for CHD. However, little research has explored or evaluated professional and parent interactions at the time of diagnosis. Future research should seek to address this gap, to inform our understanding of what parents value from professionals when receiving their child's CHD diagnosis.

The included studies predominantly involved parents who continued their pregnancy following the CHD diagnosis. Only two papers included perspectives from those who terminated the pregnancy (Carlsson & Mattsson, 2018; Leuthner et al., 2003). Previous research has provided valuable insight into decision-making regarding termination among parents receiving a prenatal diagnosis for their child (Blakeley et al., 2019). However, future research should seek to explore this specifically following a CHD diagnosis to identify possible patterns or divergences with other accounts.

The synthesis found some parents adjusted to their child's CHD diagnosis by defining a 'new normal', which can include reappraising their priorities or identity as parents. Future research could explore what the 'new normal' involves; for example, how parents' identity

may be impacted by having a child with CHD and any associated psychological entailments of this, to inform our understanding of their experiences and support needs.

## Conclusion

It has been established that parents experience distress following their child's CHD diagnosis. Findings from the current review suggest that parents may experience several phases of emotion, evolving over time and, ultimately, culminating in adjustment to the diagnosis. Parents' emotional experiences were influenced by various factors, including gender, timing of diagnosis and coping strategies employed. The review highlights the role of professionals in influencing parental wellbeing following the diagnosis, with implications for supervision in clinical practice, and further exploration of parents' preferences to improve the support following diagnosis.

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**Table 1.1**

*Search terms used for the systematic search*

| | **Domain** | **Search Terms** |
|---|---|---|
| **S** | Sample | Parent* OR mother* OR father* OR maternal OR paternal OR mum OR mom OR dad OR carer* OR caregiver* OR famil* |
| **PI** | Phenomenon of Interest | "Congenital heart disease" OR congenital heart defect* OR CHD OR heart defect* OR single ventricle |
| **D** | Design | *Not specified to ensure inclusion of all qualitative research designs* |
| **E** | Evaluation | Psych* OR anxi* OR depress* OR stress OR emotion* OR distress OR wellbeing OR "well-being" OR "quality of life" OR expe* OR "mental health" OR cope OR coping OR adjust* |
| **R** | Research Type | Qualitative OR phenomen* OR grounded theory OR thematic analysis OR content analysis OR narrative OR mixed method* |

**Figure 1.1**

*PRISMA flow diagram depicting the study selection process*

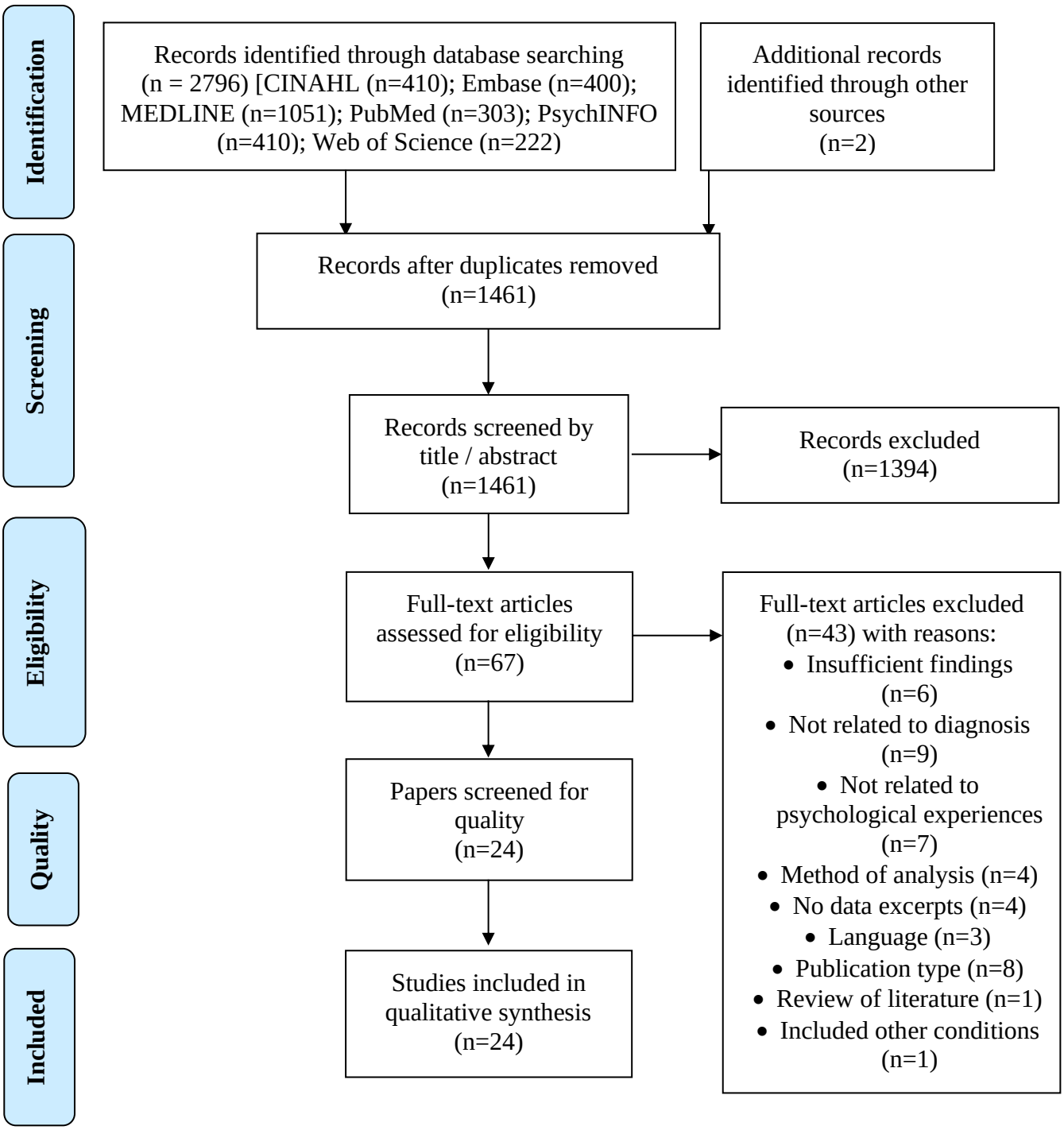


**Table 1.2**

*Characteristics of included studies in chronological order*

| | Reference and country | Research aim(s) | Sample Characteristics | | | Data collection / analysis | Relevant themes |
|---|---|---|---|---|---|---|---|
| | | | Gender and age (years) | Ethnicity | Diagnosis severity | | |
| 1 | Espinosa et al. (2021), USA | To identify emotional experiences, coping and resilience resources, and mental health treatment preferences, of pregnant and postpartum mothers receiving a CHD diagnosis. | 7 mothers, aged 27-44 | White=5<br>African American=1<br>Asian=1 | Severe | Semi-structured interview / Thematic analysis | 1. Experience of initial diagnosis<br>2. Emotional distress during pregnancy<br>3. Coping and resilience |
| 2 | Nayeri et al. (2021), Iran | To explore the meaning of being a parent of a child with CHD in Iran. | 17 mothers, 17 fathers | - | - | Semi-structured interviews / Content analysis | 1. Emotional breakdown<br>a. Denial<br>b. Shock<br>c. Sadness<br>d. Isolation<br>e. Feeling guilty<br>2. The most difficult moment in parenting life |
| 3 | Lumsden et al. (2020), UK | To explore lived experiences of parents who have children with single ventricle CHD, from diagnosis to childhood and adolescence. | 9 mothers, 3 fathers, aged 38-64 | White British=8<br>White Irish=1<br>African=2<br>Asian=1 | Severe | Semi-structured interviews / Interpretative phenomenological analysis | 1. Super parents<br>a. Parental responsibility<br>2. Accepting SVCHD and their role as parents<br>a. Timing of diagnosis |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 4 | Neubauer et al. (2020), USA | To expand understanding of the family experience of CHD from diagnosis to death. To determine whether the four transitions, described by Jones et al in adult patients and families, applies to families of children. | 10 parents, aged 29-46 | White=7<br><br>No other data reported | Severe | Semi-structured interviews / Content analysis | 1. Learning the diagnosis prenatally<br>2. Learning the postnatal diagnosis<br>3. Finding a new normal<br>4. Taking control<br>5. Realising death was likely<br>6. Communication with medical teams |
| 5 | Lee and Ahn (2020), Korea | To explore experiences and feelings of mothers facing the prognosis of their children with surgically corrected complex CHD. To present evidence for developing interventions to effectively support them. | 12 mothers, aged 40-48 | - | Severe | Semi-structured interviews / Content analysis | 1. Immense suffering<br>a. Feeling of abandonment<br>b. Anxiety with potentially losing their children<br>2. Adapting to a new life<br>a. Seeking reassurance<br>b. Trying to embrace the situation |
| 6 | Bertaud et al. (2020), UK | To explore the impact of prenatal counselling on decisions of parents of children with Hypoplastic Left Heart Syndrome (HLHS) during the antenatal period. | 8 mothers, aged 20-41 | - | Severe | Semi-structured interviews / Thematic content analysis | 1. Emotional distress and feelings of guilt<br>2. Determination to understand the condition<br>3. Value of clear explanations<br>4. Recollections of perceived pessimism<br>5. A sense of responsibility for decision-making<br>6. Maternal responsibility |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 7 | Harris et al. (2020), USA | To learn what aspects of prenatal diagnosis are particularly stressful for prospective parents. To identify potential interventions that clinicians may take to ameliorate that stress. | 16 mothers, aged 27.3-34.8; 8 fathers, aged 26-42 | White European=20 White Middle Eastern=1 Black or African American=3 Hispanic or Latino=3 | Moderate to severe | Semi-structured interviews / Thematic analysis | 1. Prenatal uncertainties<br>a. Concrete Questions<br>b. Long-term uncertainties<br>2. After the clinic visit: Identity formation |
| 8 | Hwang and Chae (2020), Korea | To describe the experience of fathers of young children with severe CHDs. | 9 fathers, mean age=39.3 | - | Severe | Semi-structured interviews / Thematic analysis | 1. Heart-breaking suffering<br>a. Unexpected family crisis related to CHD diagnosis and heart surgery<br>2. Self-control during a great struggle<br>a. Efforts to make it through to actual circumstances<br>3. Need for individualised social support |
| 9 | Williams et al. (2019), Canada | To assess parents' expectations for their child's outcome, factors they believe are important in optimising their child's resilience, barriers to optimal care and directions for future models of care. | 23 mothers, mean age=33; 3 fathers, mean age=34.9 | - | Mild to severe | Questionnaire / Thematic analysis | 1. Initial diagnosis and treatment<br>a. They (medical team) will or can save my child's life<br>b. We are not out of the woods<br>2. Overwhelming negative feelings |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 10 | Thomi et al. (2019), Switzerland | To explore parents' experiences from their new-born's CHD diagnosis through the perioperative PICU and cardiac unit stay to discharge following heart surgery. | 9 mothers, aged 30-39;<br>9 fathers, aged 31-39 | - | Severe | Semi-structured interviews / Thematic analysis | 1. Tackling a route through an unknown hospital world<br>2. Receiving the CHD diagnosis and experiencing delivery<br>3. The interplay between parents and HCPs<br>4. Being a team as a nuclear family<br>5. Concerned family members and friends |
| 11 | Im et al. (2018), Korea | To explore the pregnancy experience of women from diagnosis of foetal CHD to birth, exploring the context of accepting and deciding to continue with the pregnancy. | 12 mothers, mean age=31.5 | - | Severe | Semi-structured interviews / Grounded theory | 1. Phase of shock and pain<br>2. Phase of concern and worries<br>3. Recognition of the child as a living human being<br>4. Restructuring the pregnancy experience |
| 12 | Sood et al. (2018), USA | To understand the emotional states, stressors and supports of parents caring for a young child with CHD. | 20 mothers, 14 fathers | Non-Hispanic White=13<br>Black or African American=8<br>Hispanic/ Latino=10<br>Asian=8 | - | Semi-structured interview / Thematic analysis | 1. Emotional states<br>2. Stressors<br>3. Illness/hospital stressors |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 13 | Carlsson and Mattsson (2018), Sweden | To explore the emotional and cognitive experiences, during the time of diagnosis and decision-making, among males presented with CHD in the foetus carried by their pregnant partner. | 12 fathers, aged 20-39 | - | Moderate to severe | Semi-structured interviews / Content analysis | 1. Trying to support the partner during an emotional shock<br>2. The importance of reaching an informed and joint decision<br>3. The loss of a wanted child through an emotionally intense pregnancy termination<br>4. Ambivalent feelings of anticipation and worries about the birth |
| 14 | Woolf-King et al. (2018), USA | To explore the psychological impact of parenting a child with a critical CHD (CCHD) and the feasibility and acceptability of integrating psychological services into paediatric cardiology care. | 10 mothers, 5 fathers, mean age= 40.20 | - | Severe | Semi-structured interviews / Thematic analysis | 1. Psychological impact of parenting a child with a critical CHD<br>2. How and when to psychologically support CHD parents |
| 15 | Kim and Cha (2017), Korea | To understand experiences of fathers during the progression of their neonate's CHD. To strategize supportive intervention strategies that health professionals could use in clinical settings. | 6 fathers, aged 30-44 | - | Mild to severe | Semi-structured interviews / Descriptive phenomenology | 1. The unpreparedness to face the process of disease and the possibility of loss<br>2. Feelings involving care<br>3. Factors that hinder the confrontation of the disease and providing care<br>4. Spirituality as an imperative factor in facing the process of the disease |

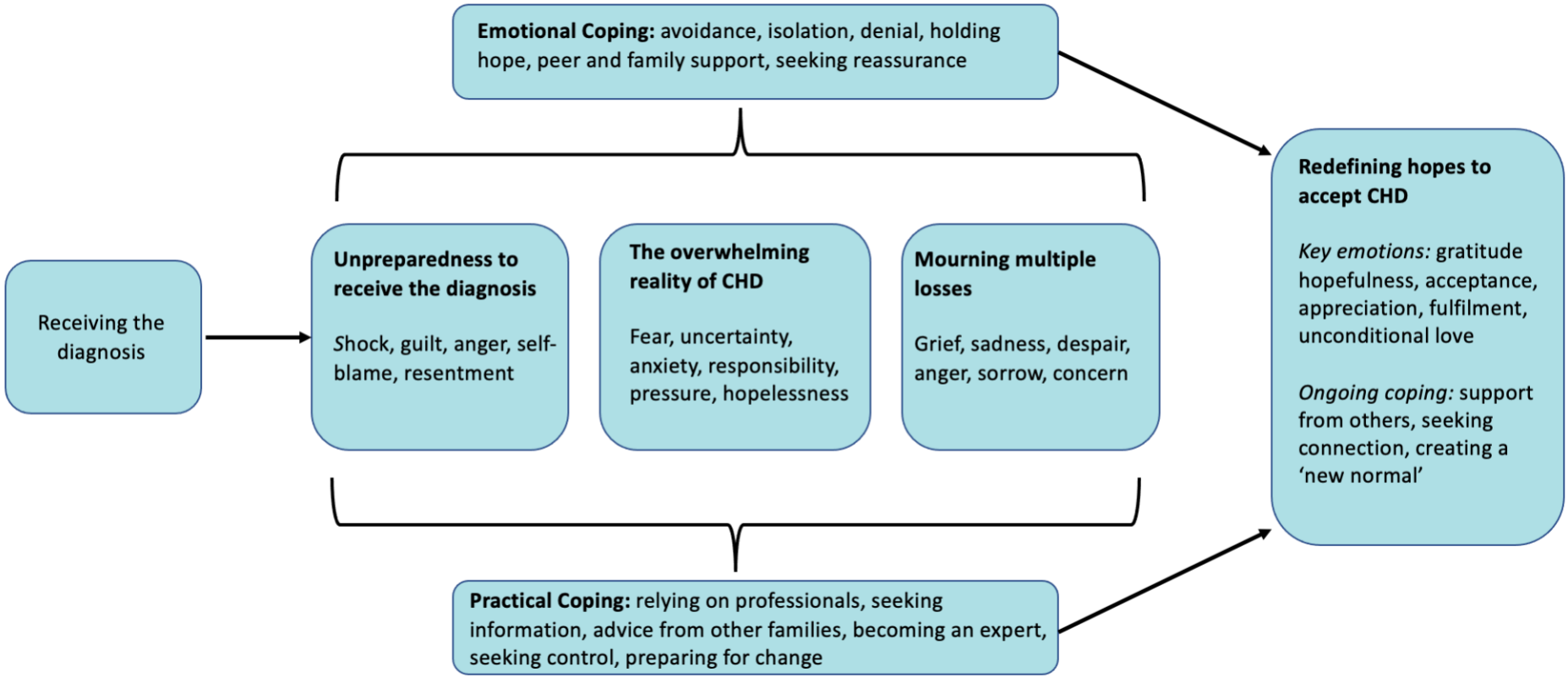
| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 16 | Barreto et al. (2016), Brazil | To understand the meaning of the experiences of parents of children with CHD. | 7 mothers, 4 fathers, aged 18-54 | - | - | Semi-structured interviews / Thematic content analysis | 1. Initially unaware of the seriousness of the disease<br>2. Falling into despair after learning about the severity of the disease<br>3. Developing feeling of guilt regarding the neonates' diagnosis<br>4. Taking full responsibility for the situation |
| 17 | Wei et al. (2016), USA | To describe parents' experiences when their child with CHD underwent heart surgery. | 10 mothers, 3 fathers, aged 19-49 | White=6 African America=5 Hispanic=2 | Mild to severe | Semi-structured interviews / Descriptive phenomenology | 1. It was a shock to hear about my child's heart defect<br>2. What did I do that caused it?<br>3. Hoping the child's heart defect would fix itself |
| 18 | McKechnie et al. (2016), USA | To explore and describe parents' caregiving motivation to manage maternal-foetal and infant health care. To examine links between parents' motivation to manage healthcare and their symptoms of distress after foetal diagnosis and after-birth treatment of CCHD. | 6 mothers, aged 23-34; 6 fathers, aged 24-31 | White=12 | Severe | Questionnaires & semi-structured interviews / Directed content analysis | 1. To determine expectations of healthcare providers<br>2. To reconcile illness and non-illness related care<br>3. To express agency as a parent |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 19 | Cantwell-Bartl and Tibballs (2015), Australia | To evaluate the psychosocial status of mothers and fathers in response to their child's diagnosis of HLHS. | 16 mothers, 13 fathers | - | Severe | Semi-structured interviews / Thematic analysis | 1. Psychosocial responses to the diagnosis<br>2. Perceptions of how the doctor delivered the news<br>3. Differences in psychosocial responses when diagnosis delivered prenatally or postnatally<br>a. The diagnosis in utero<br>b. The diagnosis after birth<br>4. Decision-making process |
| 20 | Rempel et al. (2013), Canada | To generate evidence to inform clinical practice with parents of young children with HLHS based on the perspectives of both parents and grandparents. | 15 mothers, aged 27-38;<br>10 fathers, aged 27-37 | - | Severe | Semi-structured interviews / Grounded theory | 1. Realising and adjusting to the inconceivable – "You kept on going" |
| 21 | McKechnie and Pridham (2012), USA | To examine parents' experiences of pre-birth caregiving motivations following a prenatal CCHD diagnosis | 13 mothers, 3 fathers, aged 21-29 | White=14<br>Black=1<br>Asian=1 | Severe | Semi-structured interviews / Directed content analysis | 1. Preparing heart and mind<br>2. Caregiving motivation: To relate to the baby<br>3. Caregiving motivation: To handle circumstances practically<br>4. Caregiving motivation: To manage infant medical care |

| | | | | | | | |
|---|---|---|---|---|---|---|---|
| 22 | Leuthner et al. (2003), USA | To examine the impact of an abnormal foetal echocardiogram on expectant parents' experience of pregnancy. | 9 mothers, 7 fathers, aged 28-36 | - | Severe | Focus group / Thematic content analysis | 1. Emotions<br>a. Guilt and self-blame<br>b. Fear and anxiety<br>c. Anger<br>d. Sadness or hopelessness<br>2. Coping<br>a. Attachment<br>b. Detachment<br>c. Denial<br>d. Optimism and pessimism<br>e. Privacy<br>f. Control |
| 23 | Clark and Miles (1999), USA | To explore the experience of fathers of infants newly diagnosed with CCHD during initial hospitalisation and treatment in ICU. | 8 fathers, aged 23-40 | White=6<br>African American=1<br>Asian=1 | Mild to severe | Semi-structured interviews / Content analysis | 1. Joy and sadness of becoming a father<br>2. Becoming attached while dealing with fears about the infant's outcomes<br>3. Trying to maintain control while losing control<br>4. Providing strength while hiding emotions |
| 24 | Messias et al. (1995), USA | To increase awareness and understanding among family practitioners and healthcare providers of the impact of the diagnosis of CHD on parents and family dynamics. | 7 mothers, 1 father | White=8 | - | Semi-structured interviews / Grounded theory | 1. The illusiveness of normality<br>2. The rude awakening<br>3. Managing uncertainty<br>4. New meanings |

**Figure 1.2**

*Conceptual model of themes*



**Appendix 1-A**
**Author guidelines for The British Journal of Health Psychology**

## 1. SUBMISSION

Authors should kindly note that submission implies that the content has not been published or submitted for publication elsewhere except as a brief abstract in the proceedings of a scientific meeting or symposium.

**Once the submission materials have been prepared in accordance with the Author Guidelines, manuscripts should be submitted online at http://www.editorialmanager.com/bjhp**

Click here for more details on how to use **Editorial Manager**.

All papers published in the *British Journal of Health Psychology* are eligible for Panel A: Psychology, Psychiatry and Neuroscience in the Research Excellence Framework (REF).

**Data protection:**

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**Preprint policy:**

This journal will consider for review articles previously available as preprints. Authors may also post the submitted version of a manuscript to a preprint server at any time. Authors are requested to update any pre-publication versions with a link to the final published article.

## 2. AIMS AND SCOPE

The British Journal of Health Psychology publishes original research on all aspects of psychology related to health, health-related behaviour and illness across the lifespan including:

- experimental and clinical research on aetiology
- management of acute and chronic illness
- responses to ill-health
- screening and medical procedures
- psychosocial mediators of health-related behaviours
- influence of emotion on health and health-related behaviours
- psychosocial processes relevant to disease outcomes
- psychological interventions in health and disease
- emotional and behavioural responses to ill health, screening and medical procedures
- psychological aspects of prevention

## 3. MANUSCRIPT CATEGORIES AND REQUIREMENTS

The types of paper invited are:

- papers reporting original empirical investigations, using either quantitative or qualitative methods, including reports of interventions in clinical and non-clinical populations;
- theoretical papers which report analyses on established theories in health psychology;
- we particularly welcome review papers, which should aim to provide systematic overviews, evaluations and interpretations of research in a given field of health psychology (narrative reviews will only be considered for editorials or important theoretical discourses); and
- methodological papers dealing with methodological issues of particular relevance to health psychology.

Authors who are interested in submitting papers that do not fit into these categories are advised to contact the editors who would be very happy to discuss the potential submission.

Papers describing quantitative research (including reviews with quantitative analyses) should be no more than 5000 words (excluding the abstract, reference list, tables and figures). Papers describing qualitative research (including reviews with qualitative analyses) should be no more than 6000 words (including quotes, whether in the text or in tables, but excluding the abstract, tables, figures and references). In exceptional cases the Editor retains discretion to publish papers beyond this length where the clear and concise expression of the scientific content requires greater length (e.g., explanation of a new theory or a substantially new method). Authors must contact the Editor prior to submission in such a case.

All systematic reviews must be pre-registered. The pre-registered details should be given in the methods section but blinded for peer review (i.e., 'the review was preregistered at [BLINDED]'); the details can be added at proof stage. Registration documents should be uploaded as title page files when possible, so that they are available to the Editor but not to reviewers.
Please refer to the separate guidelines for **Registered Reports**.

**COVID-19 Research**
The BJHP has received an overwhelming number of COVID-19 related submissions. We can only consider papers that are providing new and novel data on COVID-19. We particularly welcome submissions of intervention studies. Furthermore, rapid peer review for COVID-19 submissions has now ended. COVID-19 papers will now be handled alongside other standard submissions.

4. PREPARING THE SUBMISSION
**Free Format Submission**
*British Journal of Health Psychology* now offers free format submission for a simplified and streamlined submission process.
Before you submit, you will need:

- Your manuscript: this can be a single file including text, figures, and tables, or separate files – whichever you prefer. All required sections should be contained in your manuscript, including abstract, introduction, methods, results, and conclusions. Figures and tables should have legends. References may be submitted in any style or format, as long as it is consistent throughout the manuscript. If the manuscript, figures or tables are difficult for you to read, they will also be difficult for the editors and reviewers. If your manuscript is difficult to read, the editorial office may send it back to you for revision.

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To submit, login at **https://www.editorialmanager.com/bjhp/default.aspx** and create a new submission. Follow the submission steps as required and submit the manuscript.
If you are invited to revise your manuscript after peer review, the journal will also request the revised manuscript to be formatted according to journal requirements as described below.

**Revised Manuscript Submission**
Contributions must be typed in double spacing. All sheets must be numbered.
Cover letters are not mandatory; however, they may be supplied at the author's discretion. They should be pasted into the 'Comments' box in Editorial Manager.

*Parts of the Manuscript*
The manuscript should be submitted in separate files: title page; statement of contribution; main text file; figures/tables; supporting information.

*Title Page*
You may like to use **this template** for your title page. The title page should contain**:**

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- A short running title of less than 40 characters;
- The full names of the authors;
- The author's institutional affiliations where the work was conducted, with a footnote for the author's present address if different from where the work was conducted;
- Abstract;
- Keywords;
- Data availability statement (see Data Sharing and Data Accessibility Policy);
- Acknowledgments.

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Please refer to the journal's Authorship policy in the Editorial Policies and Ethical Considerations section for details on author listing eligibility. When entering the author names into Editorial Manager, the corresponding author will be asked to provide a CRediT contributor role to classify the role that each author played in creating the manuscript. Please see the **Project CRediT** website for a list of roles.

**Abstract**
For articles containing original scientific research, a structured abstract of up to 250 words should be included with the headings: Objectives, Design, Methods, Results, Conclusions. Review articles should use these headings: Purpose, Methods, Results, Conclusions. As the abstract is often the most widely visible part of your paper, it is important that it conveys succinctly all the most important features of your study. You can save words by writing short, direct sentences. Helpful hints about writing the conclusions to abstracts can be found **here**.

**Keywords**
Please provide appropriate keywords.

**Acknowledgments**
Contributions from anyone who does not meet the criteria for authorship should be listed, with permission from the contributor, in an Acknowledgments section. Financial and material support should also be mentioned. Thanks to anonymous reviewers are not appropriate.

*Statement of Contribution*
All authors are required to provide a clear summary of 'what is already known on this subject?' and 'what does this study add?'. Authors should identify existing research knowledge relating to the specific research question and give a summary of the new knowledge added by your study. Under each of these headings, please provide 2-3 (maximum) clear outcome statements (not process statements of what the paper does); the statements for 'what does this study add?' should be presented as bullet points of no more than 100 characters each. The Statement of Contribution should be a separate file.

Main Text File
As papers are double-blind peer reviewed, the main text file should not include any information that might identify the authors.
The main text file should be presented in the following order:

- Title
- Main text
- References
- Tables and figures (each complete with title and footnotes)
- Appendices (if relevant)

Supporting information should be supplied as separate files. Tables and figures can be included at the end of the main document or attached as separate files but they must be mentioned in the text.

- As papers are double-blind peer reviewed, the main text file should not include any information that might identify the authors. Please do not mention the authors' names or affiliations and always refer to any previous work in the third person.
- The journal uses British spelling; however, authors may submit using either option, as spelling of accepted papers is converted during the production process.

**References**
This journal uses APA reference style; as the journal offers Free Format submission, however, this is for information only and you do not need to format the references in your article. This will instead be taken care of by the typesetter.

**Tables**
Tables should be self-contained and complement, not duplicate, information contained in the text. They should be supplied as editable files, not pasted as images. Legends should be concise but comprehensive – the table, legend, and footnotes must be understandable without reference to the text. All abbreviations must be defined in footnotes. Footnote symbols: †, ‡, §, ¶, should be used (in that order) and *, **, *** should be reserved for P-values. Statistical measures such as SD or SEM should be identified in the headings.

**Figures**
Although authors are encouraged to send the highest-quality figures possible, for peer-review purposes, a wide variety of formats, sizes, and resolutions are accepted.
**Click here** for the basic figure requirements for figures submitted with manuscripts for initial peer review, as well as the more detailed post-acceptance figure requirements.
Legends should be concise but comprehensive – the figure and its legend must be understandable without reference to the text. Include definitions of any symbols used and define/explain all abbreviations and units of measurement.

**Supporting Information**
Supporting information is information that is not essential to the article, but provides greater depth and background. It is hosted online and appears without editing or typesetting. It may include tables, figures, videos, datasets, etc.
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- **Units of measurement:** Measurements should be given in SI or SI-derived units. Visit the Bureau International des Poids et Mesures (BIPM) website for more information about SI units.
- **Effect size:** In normal circumstances, effect size should be incorporated.
- **Numbers:** numbers under 10 are spelt out, except for: measurements with a unit (8mmol/l); age (6 weeks old), or lists with other numbers (11 dogs, 9 cats, 4 gerbils).

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Also, check out our resources for **Preparing Your Article** for general guidance and the **BPS Publish with Impact infographic** for advice on optimizing your article for search engines.

## 5. EDITORIAL POLICIES AND ETHICAL CONSIDERATIONS

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Except where otherwise stated, the journal operates a policy of anonymous (double blind) peer review. Please ensure that any information which may reveal author identity is blinded in your submission, such as institutional affiliations, geographical location or references to unpublished research. We also operate a triage process in which submissions that are out of scope or otherwise inappropriate will be rejected by the editors without external peer review. Before submitting, please read **the terms and conditions of submission** and the **declaration of competing interests**.

The Journal receives a large volume of papers to review each year, and in order to make the process as efficient as possible for authors and editors alike, all papers are initially examined by the Editors to ascertain whether the article is suitable for full peer review. In order to qualify for full review, papers must meet the following criteria:

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- the methods and/or sample size are appropriate for the questions being addressed
- research with student populations is appropriately justified
- the word count is within the stated limit for the Journal (i.e., 5000 words, or 6,000 words for qualitative papers)

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- Systematic reviews: PRISMA
- Interventions: TIDieR

We encourage authors to adhere to the APA Style Journal Article Reporting Standards for:

- Manuscripts that report primary qualitative research
- Manuscripts that report the collection and integration of qualitative and quantitative data
- Manuscripts that report new data collections regardless of research design

We also encourage authors to refer to and follow guidelines from:

- Future of Research Communications and e-Scholarship (FORCE11)
- The Gold Standard Publication Checklist from Hooijmans and colleagues
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The *British Journal of Health Psychology* recognizes the many benefits of archiving data for scientific progress. Archived data provides an indispensable resource for the scientific community, making possible future replications and secondary analyses, in addition to the importance of verifying the dependability of published research findings.

The journal expects that where possible all data supporting the results in papers published are archived in an appropriate public archive offering open access and guaranteed preservation. The archived data must allow each result in the published paper to be recreated and the analyses reported in the paper to be replicated in full to support the conclusions made. Authors are welcome to archive more than this, but not less.

All papers need to be supported by a data archiving statement and the data set must be cited in the Methods section. The paper must include a link to the repository in order that the statement can be published.

It is not necessary to make data publicly available at the point of submission, but an active link must be included in the final accepted manuscript. For authors who have pre-registered studies, please use the Registered Report link in the Author Guidelines.

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Authors are reminded that the *British Journal of Health Psychology* adheres to the ethics of scientific publication as detailed in the *Ethical principles of psychologists and code of conduct* (American Psychological Association, 2010). The Journal generally conforms to the Uniform Requirements for Manuscripts of the International Committee of Medical Journal Editors (**ICJME**) and is also a member and subscribes to the principles of the Committee on Publication Ethics (**COPE**). Authors must ensure that all research meets these ethical guidelines and affirm that the research has received permission from a stated Research Ethics Committee (REC) or Institutional Review Board (IRB), including adherence to the legal requirements of the study county.

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Once the paper is typeset, the author will receive an email notification with full instructions on how to provide proof corrections.

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*Access and Sharing*

When the article is published online:

- The author receives an email alert (if requested).
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*Author Guidelines updated April 2019*

## Appendix 1-B
## Statement of Contribution

*What is already known on this subject?*

- Parents experience shock, guilt and uncertainty following diagnosis of a congenital anomaly in their child
- Interactions with health professionals can influence parents' emotional experiences of receiving this diagnosis.
- Parents are at increased risk of experiencing mental health difficulties when they receive their child's diagnosis of congenital heart disease (CHD).

*What does this study add?*

- Following the diagnosis, parents experienced phases of emotion, including shock, fear, grief and acceptance.
- Experiences differed between mothers and fathers, and those receiving a prenatal compared to postnatal diagnosis.
- Parents struggled to reconcile the joy of pregnancy with CHD. They valued clinicians' compassion.

**Appendix 1-C**
**Example search strategy for PubMed**

| Item | Search terms | Location | Results |
|---|---|---|---|
| S1 | Parent* OR mother* OR father* OR maternal OR paternal OR mum OR dad OR carer* OR caregiver* OR famil* OR mom | Title or abstract | 2,181,176 |
| S2 | "Parents"[MeSH] OR "Fathers"[MeSH] OR "Mothers"[MeSH] | Title or abstract | 132,055 |
| S3 | "Congenital heart disease" OR congenital heart defect* OR CHD OR heart defect* OR single ventricle | Title or abstract | 69,943 |
| S4 | "Heart Defects, Congenital"[MeSH] | Title or abstract | 164,046 |
| S5 | heart N5 (disease OR defect)) | Title or abstract | 3,748 |
| S6 | Psych* OR anxi* OR depress* OR stress OR emotion* OR distress OR wellbeing OR "well-being" OR "quality of life" OR expe* OR "mental health" OR cope OR coping OR adjust* | Title or abstract | 7,141,164 |
| S7 | ("Mental Health"[Mesh]) OR "Stress, Psychological"[Mesh] | Title or abstract | 194,323 |
| S8 | Qualitative OR phenomen* OR grounded theory OR thematic analysis OR content analysis OR narrative OR mixed method* | Title or abstract | 727,187 |
| S9 | ("Qualitative Research"[MeSH]) | Title or abstract | 74,205 |
| S10 | S1 OR S2 | | 2,198,719 |
| S11 | S3 OR S4 OR S5 | | 203,954 |
| S12 | S6 OR S7 | | 7,170,603 |
| S13 | S8 OR S9 | | 739,220 |
| S14 | S10 AND S11 AND S12 AND S13 | | 303 |

**Appendix 1-D**
**Quality appraisal**

| Study | Clear aim? | Appropriate methodology? | Research design | Sampling | Data collection | Reflexivity | Ethical issues | Data analysis | Findings | Research value | Total |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 1 | Yes | Yes | 2 | 2 | 3 | 1 | 3 | 2 | 3 | 3 | **19** |
| 2 | Yes | Yes | 3 | 1 | 3 | 1 | 2 | 2 | 2 | 2 | **17** |
| 3 | Yes | Yes | 3 | 3 | 3 | 3 | 3 | 3 | 3 | 3 | **24** |
| 4 | Yes | Yes | 2 | 2 | 2 | 1 | 3 | 2 | 2 | 2 | **16** |
| 5 | Yes | Yes | 3 | 2 | 3 | 1 | 3 | 3 | 3 | 2 | **20** |
| 6 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 3 | 3 | 2 | **20** |
| 7 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 3 | 3 | 2 | **20** |
| 8 | Yes | Yes | 3 | 3 | 3 | 2 | 3 | 2 | 3 | 3 | **22** |
| 9 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 2 | 3 | 2 | **19** |
| 10 | Yes | Yes | 3 | 3 | 3 | 1 | 3 | 3 | 3 | 3 | **22** |
| 11 | Yes | Yes | 3 | 3 | 3 | 1 | 3 | 3 | 2 | 3 | **21** |
| 12 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 3 | 3 | 2 | **19** |
| 13 | Yes | Yes | 3 | 3 | 3 | 2 | 3 | 3 | 3 | 3 | **23** |
| 14 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 2 | 3 | 2 | **19** |
| 15 | Yes | Yes | 3 | 2 | 3 | 1 | 2 | 3 | 3 | 2 | **19** |
| 16 | Yes | Yes | 2 | 2 | 3 | 1 | 2 | 3 | 2 | 1 | **16** |
| 17 | Yes | Yes | 3 | 3 | 3 | 1 | 3 | 3 | 3 | 3 | **22** |
| 18 | Yes | Yes | 2 | 2 | 3 | 1 | 3 | 2 | 3 | 3 | **19** |
| 19 | Yes | Yes | 3 | 3 | 3 | 1 | 3 | 2 | 2 | 2 | **18** |
| 20 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 3 | 3 | 3 | **21** |
| 21 | Yes | Yes | 2 | 3 | 3 | 1 | 3 | 2 | 2 | 2 | **18** |
| 22 | Yes | Yes | 2 | 2 | 3 | 1 | 3 | 2 | 2 | 2 | **17** |
| 23 | Yes | Yes | 2 | 2 | 3 | 1 | 2 | 2 | 3 | 2 | **17** |

| | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 24 | Yes | Yes | 2 | 2 | 3 | 1 | 1 | 2 | 1 | 2 | **14** |

1 = weak evidence or justification; 2 = moderate evidence or justification but not fully explained; 3 = strong evidence or justification

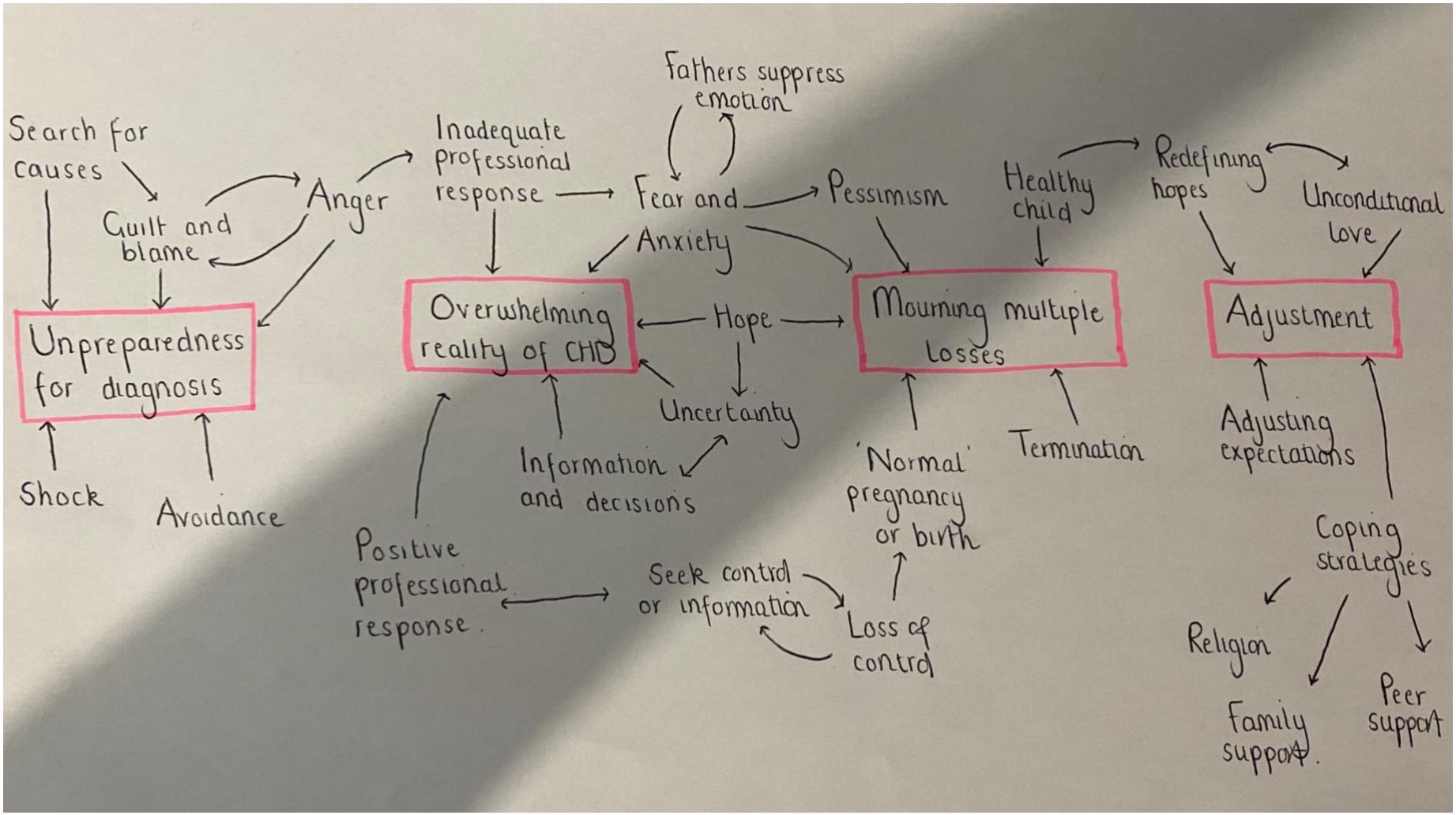
**Appendix 1-E**
**Example of transformation of line-by-line coding to descriptive and analytical themes**

**Theme 3: Mourning multiple losses**

| Line-by-line codes | Descriptive Themes | Analytical Theme |
|---|---|---|
| • Diagnosis interfered with expectations for pregnancy<br>• Fear that nurses would know the baby better than they would<br>• Ruminating on inability to have a 'normal' birth<br>• Mothers adjusting to increased prenatal monitoring<br>• Diagnosis interfered with joy at becoming a parent<br>• Pregnancy became serious and worrying instead of happy<br>• Juxtaposition of hope and despair<br>• Prenatal diagnosis facilitated feeling prepared for a different birth<br>• Prenatal diagnosis created time for rumination and worry<br>• Sadness and sorrow at diagnosis<br>• Unpreparedness to navigate medical world | Loss of a normal pregnancy and birth experience | Mourning multiple losses<br>a) Loss of a 'normal' pregnancy<br>b) Loss of a healthy child and envisioned future |
| • Termination led to concrete loss of the child<br>• Termination likened to an 'execution'<br>• Diagnosis means feeling the world collapse<br>• Fear of the child dying<br>• Hoping that the child would be healthy<br>• Loss of a healthy, normal child<br>• Sadness and sorrow at diagnosis<br>• Stigma of an 'imperfect' child | Loss of a healthy / perfect child | |
| • Fear they and their child would never live a normal life<br>• Feeling unprepared to parent the child<br>• Hoping the child could live a normal life, despite CHD<br>• Diagnosis interfered with plans and expectations for parenthood<br>• Loss of envisioned parenthood | Loss of envisioned parenthood | |

| | |
|---|---|
| • Pessimism about the future<br>• Anxiety about fulfilling parental roles | |
| • Being strong as a man's duty<br>• Expressing emotions alone | Expressions of grief different between mothers and fathers |

**Appendix 1-F**
**Development of analytical themes**



**Appendix 1-G**
**Matrix of included studies and identified themes**

| Study | Theme 1: Unpreparedness for the diagnosis | | | Theme 2: The overwhelming reality of CHD | | | Theme 3: Mourning multiple losses | | Theme 4: Redefining hopes to reach acceptance of CHD | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| | Shock | Blame | Avoidance | Information & decisions | Uncertainty & fear | Coping | 'Normal' pregnancy | Healthy child | Coping strategies | Adjusting expectations | Redefining hopes |
| 1 | Y | Y | - | Y | Y | - | - | Y | - | - | - |
| 2 | Y | Y | Y | Y | - | - | - | - | - | - | - |
| 3 | - | Y | - | - | - | - | - | - | - | - | - |
| 4 | Y | - | - | Y | Y | - | Y | Y | - | Y | Y |
| 5 | - | Y | - | - | Y | Y | - | - | Y | Y | Y |
| 6 | - | Y | - | Y | - | Y | - | - | Y | - | - |
| 7 | - | Y | - | Y | Y | - | Y | Y | Y | - | Y |
| 8 | Y | - | - | - | Y | Y | - | Y | - | - | - |
| 9 | - | - | - | - | Y | Y | - | Y | - | - | Y |
| 10 | Y | Y | - | Y | Y | Y | Y | Y | - | - | - |
| 11 | Y | Y | Y | Y | Y | Y | - | - | - | Y | - |
| 12 | Y | Y | - | - | - | - | - | - | - | - | - |
| 13 | Y | - | Y | Y | Y | - | Y | Y | - | - | - |
| 14 | Y | Y | - | - | - | - | - | Y | Y | - | - |
| 15 | Y | Y | Y | - | - | - | - | - | - | - | - |
| 16 | Y | Y | Y | - | Y | Y | - | Y | - | - | - |
| 17 | Y | Y | - | - | Y | Y | - | - | - | - | Y |
| 18 | - | - | - | Y | Y | Y | Y | Y | - | Y | - |
| 19 | Y | - | Y | Y | - | Y | Y | - | Y | - | - |
| 20 | - | - | - | - | - | - | Y | - | - | Y | - |
| 21 | - | Y | Y | Y | Y | Y | Y | Y | Y | - | Y |
| 22 | - | Y | Y | Y | Y | Y | Y | Y | - | Y | - |
| 23 | - | - | - | - | Y | Y | - | Y | - | - | - |

| | | | | | | | | | | | |
|---|---|---|---|---|---|---|---|---|---|---|---|
| 24 | Y | - | Y | - | Y | Y | - | Y | Y | Y | Y |

**Section 2: Research Paper**

# Parenting a child with single ventricle congenital heart disease: Mothers' experiences of role and identity

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**Abstract**

**Objectives:** The study aimed to explore how having a child with single ventricle congenital heart disease (SVCHD) affected mothers' experiences of their parental role and identity.

**Design:** This was a qualitative, semi-structured interview study.

**Methods:** Eight mothers whose children had undergone the Fontan surgical procedure were recruited via social media. Interviews were completed using Microsoft Teams and audio-recorded, then transcribed and analysed using Interpretative Phenomenological Analysis.

**Results:** Four themes were identified: 1) being a "heart mum", 2) managing competing roles: "you have to wear lots of different hats all at the same time", with subthemes a) promoting normality vs. protecting the child and b) mothering vs. nursing roles, 3) loss and regaining of identity and 4) relinquishing control and letting go of caring roles.

**Conclusions:** Parenting a child with SVCHD presented significant challenges to mothers' parental role and identity, which they managed in various ways. There are implications for health services to support mothers with their psychological wellbeing, managing nursing roles and their child's transition to adulthood.

***Key words: Congenital Heart Disease, Parenting, Identity, Role, Interpretative Phenomenological Analysis***

# Parenting a child with single ventricle congenital heart disease: Mothers' experiences of role and identity

Congenital heart disease (CHD) is a structural heart defect present at birth and the most common congenital anomaly. The estimated global prevalence is 1%, with higher rates in low socio-demographic areas, such as the Middle East (Zimmerman et al., 2020). Although survival rates have improved, CHD severity ranges from mild to severe, so treatment and prognosis vary. For instance, single ventricle CHD (SVCHD), in which part of the heart is unformed, requires several life-saving surgeries, culminating with the Fontan surgery at approximately age four. These children require ongoing care, such as medication and catheter procedures (Kaulitz & Hofbeck, 2005). However, treatment for SVCHD remains palliative and a meta-analysis of 16 studies found only 59.8% survive to five years of age (Best & Rankin, 2016).

Reviews of quantitative and qualitative literature suggest that children with CHD and their families are vulnerable to psychosocial distress because they experience additional challenges, such as hospitalisation (Abda et al., 2019; Lumsden et al., 2019; Soulvie et al., 2012; Wei et al., 2015). However, Jackson et al. (2015) found individual factors, including financial stability and familial cohesion, have greater impacts on wellbeing than CHD itself. Given the prevalence of distress among these children and their families, several countries, including the UK (NHS England, 2016) and Australia (Department of Health, 2019), recommend that psychological support is available at any stage of care.

Parental mental health can affect cognitive and emotional development in children with health conditions (Leeman et al., 2016), so research has explored psychological wellbeing among parents of children with CHD. These parents are more likely to experience anxiety and depression (Alkan et al., 2017; Lawoko & Soares, 2002) and post-traumatic stress (Kolaitis et al., 2017; Woolf-King et al., 2017) than parents of healthy children. In

qualitative studies, parents of children with CHD describe isolation, guilt and fear throughout their child's life (Lumsden et al., 2020). Findings are consistent across cultures (Im et al., 2018; Nakazuru et al., 2017) and may be explained by the additional challenges faced, such as making important medical decisions (Rempel et al., 2004) or adapting to necessary financial or lifestyle changes (Connor et al., 2010). Indeed, stages in which such challenges are most acute are associated with increased distress, including diagnosis (Bratt et al., 2019), hospitalisation (Lisanti et al., 2017; Simeone et al., 2018) and surgery (Harvey et al., 2013; Wei et al., 2016).

In contrast to the above patterns of findings, some studies found no difference in levels of distress between parents of children with CHD and normative samples (Fischer et al., 2012; Spijkerboer et al., 2007; Vrijmoet-Wiersma et al., 2009). However, the authors acknowledged that results may have been affected by difficulties with measuring distress or lack of comparison groups. Nonetheless, the findings may simply suggest that some parents are able to psychologically adapt to their child's condition. For example, parents of children with SVCHD interviewed by Lumsden et al. (2020) reported establishing a sense of normality once they had adjusted to changes the condition brought to their parental role.

A person's understanding of, and adjustment to, their parental role and identity can affect their psychological wellbeing and, in turn, that of their child (Cast, 2004; Fadjukoff et al., 2016). Definitions of role and identity are debated but understanding can be aided by identity theory. Identity is considered a set of meanings, according to the roles a person adopts and groups they belong to, that shape understanding of who they are (Stets & Serpe, 2013). Individuals are thought to have multiple identities (e.g., partner, worker), organised into a hierarchy (Stryker & Burke, 2000). Conversely, roles are behaviours or responsibilities that inform an identity (Ihinger-Tallman et al., 1995).

Being a parent prompts renegotiation of an individual's identity and roles (Cast, 2004). Although experiences differ, for example between genders (Katz-Wise et al., 2010), parental identity is usually prioritised and includes roles such as nurturer, protector and disciplinarian (Ihinger-Tallman et al., 1995). Enacting these roles can verify an individual's appraisal of their parental identity, promoting psychological wellbeing. However, parents also report feeling inadequate in their perceived roles or challenged by competing demands of other identities, which can cause distress (de Montigny & Lacharité, 2005; Petch & Halford, 2008).

The negotiation of parental role and identity is also affected by a child's wellbeing. Systematic literature reviews by Shudy et al. (2006) and Abela et al. (2020) found parental role, identity and wellbeing were acutely challenged when the child was critically ill. Similar findings are reported among parents of children with chronic illnesses (Kirk et al., 2005; McKenzie & Curle, 2012; Nicholas, 2017; Young et al., 2002) and disabilities (Pertriwskyj et al., 2016). Diffin et al. (2016) and Lisanti et al. (2021) also found parental role alteration in intensive care environments (e.g., being unable to hold the child) significantly increased distress among parents of children with CHD.

While qualitative research has not explored identity among parents of children with CHD, it has offered limited consideration of parental roles relative to wider experiences. For example, feeling stripped of parental roles during hospitalisation (Simeone et al., 2018), as a father (Gower et al., 2017), or struggling to be both parent and nurse following hospital discharge (Elliott et al., 2021; Gaskin, 2018; Harvey et al., 2013). These experiences were associated with psychological distress. Two recent studies considered parental role in more detail. Fisk et al. (2022) described two categories of parental roles in the cardiac intensive care unit: decision-maker and advocate, as well as providing emotional and physical support. There was little discussion regarding the psychological implications of these roles, but they

were reported to be valued. Considering the whole parental experience, Lumsden et al. (2020) identified parental roles to promote normality, by treating their child similar to peers, and protect their child through hypervigilance. These roles were associated with pride and fear, respectively. Experiences of parental role, therefore, seem to influence psychological wellbeing among parents of children with CHD.

The lack of qualitative research into parental role and identity among parents of children with CHD means there is limited understanding of their experiences of these concepts. Thus, the current study aimed to explore the lived experience of parental role and identity among parents of children with SVCHD. This qualitative study focused on SVCHD because it has been associated with more acute challenges that may affect parental role and identity, such as increased caring responsibilities (Solberg et al., 2012). The research question was: 'what is the lived experience of parental role and identity for parents of children with SVCHD?'

## Methods

### Design

The study used qualitative methodology, namely semi-structured interviews, because the aim was to gain qualitative information about participants' experiences. Data was analysed using Interpretative Phenomenological Analysis (IPA), which has theoretical underpinnings in idiography, phenomenology and hermeneutics (Smith, 1996). Thus, IPA corresponded with the research aim because it is concerned with participants' lived experiences, and the associated psychological entailments and meanings given to these (Smith et al., 2009). The idiographic nature of IPA also allows exploration of patterns and divergences within and across participant accounts (Murray & Wilde, 2020). IPA has also proven insightful in research considering identity (Smith, 2004) and health (Smith, 2011),

and has been used in similar research into experiences of having a child with SVCHD (Lumsden et al., 2020).

## Ethical Approval

The study was approved by the Lancaster University Faculty of Health and Medicine ethics committee, following the submission of an application form and research protocol (reference: FHMREC20023). The ethics application and approval are located in Section 4.

## Sampling and Participants

The idiographic focus of IPA means it is recommended to purposively establish a small, homogenous sample with similar characteristics or experiences, to best support the researcher's understanding of these (Murray & Wilde, 2020). Participants had to reside in the UK and have a child age 25 years or younger with SVCHD who had their completed Fontan surgery at least 6 months prior to interview. The rationale for this criterion was: 1) to promote homogeneity among the sample and 2) to allow exploration of the complete surgical journey, whilst acknowledging that the immediate post-surgery period can be particularly distressing (Harvey et al., 2013). Participants also had to be proficient in the English language. Parents were ineligible to take part if their child had undergone heart transplant or was hospitalised at the time of interview, because these experiences have a separate nature from those being explored (Simeone et al., 2018; Woolf-King et al., 2017).

## Procedure

The study was advertised via social media created for the research, and the social media, email lists, websites and newsletters of UK heart charities. The advertisement included an electronic participant information sheet and individuals were invited to email the

lead researcher to discuss participation. Prior to arranging an interview, participants completed an 'expression of interest' form to ensure they met inclusion criteria. This included demographic information (age, sex, child's age, time since Fontan surgery), which was confirmed and added to (ethnicity, employment status, marital status) verbally at the beginning of each interview.

Interviews took place remotely between April and October 2021, due to the ongoing Covid-19 pandemic, and lasted between 72 and 117 minutes (mean=86.63, SD=14.85). Following ethics committee advice, Microsoft Teams was used for all interviews. Verbal consent was confirmed and audio-recorded prior to the interview, with each item on the consent form read out by the lead researcher and verbally agreed to by the participant. Interviews were guided by a topic guide and recorded using a dictation device. Following the interview, participants were offered debriefing information containing details of further support and asked if they would like to receive a summary of the research. All research materials are included in Section 4.

**Data Collection**

Semi-structured interviews allow the researcher to maintain focus on the research question, whilst providing opportunities for participants to recall their experiences in detail (Smith et al., 2009). Thus, following review of the literature, input from the research team and consultation with a national heart charity, an interview topic guide was developed. The topic guide aimed to explore parents' understanding of their role and identity relative to having a child with SVCHD, including any associated impacts upon their psychological wellbeing. It was reviewed following each interview to consider necessary revisions and was adjusted once to incorporate consideration of the difference between mothers' and fathers'

experiences when parenting a child with SVCHD, because the first participant placed importance on this.

## Data Analysis

The lead researcher transcribed interview recordings verbatim, using pseudonyms to maintain confidentiality. Data were analysed using IPA, as described by Murray and Wilde (2020) (Appendix 2-B). To begin, the lead researcher became familiar with the data by reading and re-reading each transcript. Notations were made, and initial codes generated, regarding participants' experiences of their identity and roles when parenting a child with SVCHD (see example in Appendix 2-C). For each transcript, initial codes were grouped into themes, summarised narratively and titled. Themes were then compared across participants' accounts and, again, grouped with an interpretative summary written about each. The themes were amended (e.g., combining multiple themes together) through discussion with the research team to form the final themes which encompassed the lead researcher's understanding of experiences of role and identity when parenting a child with SVCHD.

## Credibility

To support the credibility of analysis, an audit trail was kept for each interview (see example for participant 1, Emma, in appendix 2-D). The research team included an experienced IPA researcher (CM), so the audit trail for Emma was shared and discussed for feedback regarding the interpretations made before analysis of subsequent interviews. The same process of sharing, discussion and feedback was completed before merging themes across transcripts. Furthermore, each theme was represented by data extracts from at least half of the participants, consistent with guidance regarding the presentation of data in IPA studies (Smith, 2011).

### Reflexivity

Hermeneutics is concerned with interpretation and is central to IPA. IPA recognises a process of 'double hermeneutics', in which participants try to make sense of and articulate their experiences and the researcher, in turn, tries to make sense of and interpret these. Thus, it is important to recognise and 'bracket' the researcher's pre-existing assumptions or beliefs to prevent undue influence on the analysis (Smith et al., 2009). This was achieved using a reflective diary and discussing reflections in supervision. For example, the lead researcher is a white, working-class woman who has worked psychologically with children and families in a Cardiology team at a tertiary children's hospital. This experience and its influence on expectations of the research were discussed regularly throughout.

## Results

Twelve individuals responded to the study advertisement. Two did not meet inclusion criteria and two did not respond to correspondence to arrange an interview. Thus, eight individuals participated. Demographic information is located in Table 2.1. Participants were all mothers aged 36-57 years (mean=47.25, SD=6.320). All were White and six were employed. Six participants were married, one was single, and one was separated. Their children were aged 4-22 years (mean=16.75, SD=5.19) and their child's Fontan surgery had been between 9 months and 18 years prior (mean=12.34, SD=5.02).

The analysis generated four themes: 1) being a "heart mum", 2) managing competing roles: "you have to wear lots of different hats all at the same time", with subthemes a) promoting normality vs. protecting the child and b) mothering vs. nursing roles, 3) loss and regaining of identity, and 4) relinquishing control and letting go of caring roles. Figure 2.1 illustrates a conceptual model of themes, and Appendix 2-E details how individual

participants contributed to these. Figure 2.2 illustrates how parental role and identity appeared to change over time. A summary of each theme is detailed below.

[INSERT TABLE 2.1, FIGURE 2.1 AND FIGURE 2.2 HERE]

**Theme 1: Being a "heart mum"**

The diagnosis disrupted expectations of parenthood and, informed by caring roles they adopted, participants developed a parental identity of "heart mum", which was characterised by guilt and self-criticism: "You feel like you're not doing a good job. It affects your sense of self, because you have all these ideas about being a parent that just aren't possible" (Elise). Kerry added that medical roles left less time for her other children: "The other two are like: "that's not fair" and "it's always about [child]", so I do feel like a bad mum". Thus, there was a sense of resentment and injustice about being a "heart mum", particularly for Lisa, who experienced fertility difficulties and spent longer envisioning parenthood: "I certainly remember feeling jealous of parents who have healthy children. I felt I'd been robbed of something".

Most considered the "heart mum" identity to be lifelong. However, Lisa disconnected from it when her son became unexpectedly healthier aged three, suggesting acute illness is key: "If he'd been much more unwell then maybe I'd still consider myself a heart mum". Indeed, risk of harm differentiated this from a typical parental identity: "You couldn't just be 'mum' when you're waking up and your first thought is: 'is my child alive?'" (Lisa). Even compared to her children with other CHD types, Holly noted differences: "I suppose, how I feel about myself as a mother is really different. I was nervous and felt all this responsibility". The fear and responsibility inherent to being a "heart mum" seemed to limit the enjoyment participants took from this identity: "I tried hard to be upbeat and positive, to

enjoy my child and have a normal experience, but those additional responsibilities round every corner just knock you down" (Emma).

The medical risk and responsibilities of SVCHD also made feeling different central to the "heart mum" identity, seeming to create sadness, frustration and isolation. Holly felt unable to relate to others: "It's hard not to feel like the jealous parent, or frustrated or whatever, because their biggest worry is a tiny little one compared to mine". Likewise, Sophie recalled exasperation when other parents' worries seemed trivial compared to the life-threatening nature of SVCHD:

> Mums were talking about organic ketchup. I said, 'well wouldn't you just want your child fed?' and that didn't get a good response… I thought, how privileged you must be to make an informed choice about what your child eats.

Similar feelings were created by complimentary judgements, suggesting difference specifically affects wellbeing: "People seem to think: 'how does she do it?' but I just do what I have to, to protect [child]… It can be really lonely when people think you're a super-mum" (Lisa).

Some mothers positively appraised the "heart mum" identity by finding meaning in difference: "I've coped with my faith, you know, that we were given him for a reason" (Jen). Others became disillusioned by feeling judged and excluded, so tried to avoid or hide the identity. For instance, Sophie responded to being labelled "freakish" and excluded from activities: "I remember agreeing with those mundane worries to hide that I was different". Over time, most developed pride in their achievements as a "heart mum": "I'd cover her up. I didn't want people seeing it [scar]… As she got older, though, I took that T-shirt off and I was like 'no, I'm proud of that scar. That scar means you're alive'" (Kerry). This acceptance of the identity seemed to facilitate positive outcomes. For example, Elise embraced peer

support that she previously avoided: "I now prefer being with parents with a similar outlook, who know their child is precious".

## Theme 2: Managing competing roles: "You have to wear lots of different hats all at the same time"

### *Promoting normality vs. protecting the child*

Trying to balance multiple additional roles was stressful and exhausting: "Being the parent of a heart child, you have to wear lots of different hats all at the same time" (Emma). Kerry summarised a central dilemma: "That's difficult, trying to balance letting her be normal with keeping her safe". The risk of early death created urgency to facilitate normal experiences: "Making sure she'd experienced things like putting her feet on the beach, because we didn't know how long we'd have her for" (Sophie). After children survived, mothers longed to promote normality to minimise the impact of SVCHD but feared associated risks, so adopted protective parenting styles: "Her condition made us more cautious with all of them" (Clare). Mothers experienced roles to protect more intensely for their children with SVCHD and described hypervigilance: "I wrapped her up in cotton wool, no one was gonna go near her or hurt her. That's my special baby" (Kerry). There was a sense of guilt about this: "I feel like bad cop when there's a new activity and I say no because of her condition" (Emma). Kerry used emotional resilience to navigate the dilemma, recalling allowing her child on a trampoline: "I was terrified, but she needed a bit of normality. I had to hold that emotion inside". Most participants also adapted 'typical' activities to remain reassured about safety: "They wanted to go camping but she wouldn't have managed that… so we'd put sleeping bags in the living room" (Clare). This required additional planning, but mothers accepted the burden because it allowed them to feel the relief normality provided.

Sophie translated medical notes for holidays: "I realised this was a lot of effort for a week, but it allowed us to have the normal experience".

Participants were unwilling to compromise protecting their child through advocacy (e.g., seeking support or challenging professionals), because they felt others lacked understanding: "She needed me to advocate more because often her condition was hidden, so there'd be things she couldn't do but people wouldn't understand" (Emma). The hidden nature of SVCHD appeared to make advocacy difficult because mothers were labelled 'overprotective'. Even medical professionals, who understood the condition, could be experienced as patronising, creating sadness and anger: "Everyone saw me in that way of questioning them or being overly protective and I felt really isolated" (Holly). Thus, mothers felt they had to be 'gobby' or 'pushy'. For some, this created pride: "If I didn't [advocate] she wouldn't be here and she wouldn't have the things she needs" (Kerry). Others felt it contrasted with their personality, creating embarrassment and anxiety: "I was quite shy, I wouldn't have spoke up about anything… but with a heart child you really have to… it did make me feel sick" (Clare). Again, these mothers exercised emotional resilience to prioritise their child's needs, as the risks of SVCHD made advocacy imperative.

### *Mothering vs. nursing roles*

During hospitalisation, participants felt helpless and took pride in mothering roles, which provided normality: "I had no role in intensive care, but anything I could do, I did. I put a hat and some booties on her, just normal mum stuff" (Kerry). Conversely, nursing roles seemed overwhelming. Clare tube-fed her child: "You have to make sure it's in right or it could go in her lung. I remember thinking "oh god" and feeling under so much pressure". Despite having older children with CHD, Holly added: "I take on those roles of being her nurse much more intensely". Indeed, there was a sense that nursing roles were informed by

guilt about other parents' losses to SVCHD: "Not everyone was as fortunate as we were to take our child home. It put a lot of added pressure on" (Lisa). Participants prioritised nursing roles to protect their child but seemed to grieve 'normal' mothering experiences: "So much was doing stuff to her that the time I spent with her wasn't mother-daughter" (Sophie). Most coped pragmatically and detached from their emotions. Holly delayed accessing support due to fear of "falling apart": "You can't really think about it, you just get on with it". For some, there was a sense of mothering through it all. Sophie created opportunities to be 'mum' wherever possible: "It felt like reading a book with her was such a big deal". Similarly, Jen described managing medical procedures: "It was like: 'hey now, you just look at me and lean into me', being comforting and supportive as his mum".

However, frustration that nursing roles fell to mothers, not fathers, remained: "He didn't do all the researching and worrying I did… He was able to get on with things because I took that on" (Jen). Nonetheless, having control over nursing roles seemed to provide reassurance: "He'd avoid the difficulties… It could be frustrating, but I also wanted to do it. I wanted to know all of what was going on" (Elise). Over time, becoming practised in medical care allowed assimilation of nursing roles into daily life: "We got used to it, like giving her Weetabix in the morning, medicine was the same" (Clare). This appeared to facilitate acceptance, as mothers realised a balance of roles was possible.

**Theme 3: Loss and regaining of identity**

In addition to the "heart mum" identity, mothers' personal identity was impacted by parenting a child with SVCHD. Whilst being a "heart mum" involved various psychological responses (e.g., guilt, resentment, anxiety), this third theme encompasses additional experiences of losing qualities and being unable to take part in activities that were important to mothers' sense of self. Participants felt distressed that they were unable to maintain

previously valued identities (e.g., friend, worker), which were overshadowed by the "heart mum" identity and associated medical roles:

> I wanted to build friendships, but I couldn't because I'd have to keep dipping out of those circles to spend time in hospital… I couldn't go back to work because I needed to look after her and I just felt consumed by it all (Emma).

Some expressed self-criticism about changes to their identity: "I didn't want to be this person, living off the state. I had bad opinions about that" (Kerry). Jen added: "I became a more serious person because my anxiety levels were so high". There was a sense of helplessness when others reinforced this: "I didn't have time for anything else [outside medical care], which made me feel quite low… Friends would say I wasn't the person I was before. Work colleagues also said that, but I just had nothing left to give" (Elise). Thus, parenting a child with SVCHD appeared to leave mothers feeling powerless to retain a personal identity.

Most understood identity loss as a temporary necessity, due to fear of the potential consequences if being "heart mum" was not their sole priority: "I never had the confidence to go off and do something for myself 'cause I always had to be around and worried what would happen if I wasn't" (Jen). However, making an active choice to prioritise medical responsibilities seemed to restore a sense of control: "I've accepted that, for the time being, everything is on hold to focus on this, and I will go and be me at some point" (Sophie). Nonetheless, there was jealousy that fathers did not experience this: "He was able to go to work and keep that part of him… I always used to be jealous when he would go on little work trips" (Jen). Conversely, Lisa's experience of almost losing her child prompted a new outlook on life. As her child became healthier at a younger age and had fewer medical needs, she felt able to act on this and retain her sense of self: "I was able to realise that there's a lot more to

my life than just being a 'heart mum'. That was helped by me working full-time and doing other hobbies".

The process of regaining a personal identity evolved over time. Holly, whose child was younger, had not reached this stage: "I'm still finding my way back to me". Most only felt able to take time away from nursing roles when their child became healthier, which was met with relief and fulfilment: "I literally couldn't stop or else something really bad could happen to him. Whereas now, I've started to find myself again. I do yoga and a lot of exercise, being sporty is a real part of who I am" (Elise). This suggests participants' personal identity and that of "heart mum" competed against one another. However, over time, the two appeared to become integrated into participants' overall sense of who they were: "Those parts of my identity ["heart mum" and friend] aren't separate anymore" (Clare). Indeed, some found the experience of extreme loss eventually prompted greater connection to their identity: "I had more confidence to be my authentic self, to say who I was and what I believed in" (Emma). Similarly, Elise stated: "I feel like I've gone from being hemmed in by it all to being this larger person, so I've grown from it".

In summary, the particular circumstances of being a "heart mum" meant mothers felt unable to maintain previously valued identities, such as friend or worker, and became people they didn't want to be (e.g., serious). Some accepted that their personal identity needed to be on hold during a period of intense medical responsibility and regained their sense of self over time, through valued activities.

**Theme 4: Relinquishing control and letting go of caring roles**

Over time, participants recognised a need to let go of caring responsibilities and pass these onto their child: "As the years went on, I've tried to let him take charge" (Jen). Indeed, only Holly, whose child was much younger, did not discuss this. Clare described the process

of letting go: "It's like a dip in a rollercoaster, we've took them [roles] all on and been doing them to then having to just let them go". Therefore, in contrast to typical parenthood, the lengthy process of adjusting to additional responsibilities associated with SVCHD created an unpreparedness for relinquishing control over their child's care.

Letting go left mothers to reflect on their life without additional caring roles. For some, there was immediate relief: "I don't feel there's much of a nursing role anymore… That's a pressure off me" (Lisa). However, a sense of ambivalence emerged when they understood that letting go meant losing control over their child's care, which had become a coping strategy:

> I find it hard with the stages of independence… because I'm not in control of them. I know that's a problem, but I've lived so anxiously and on edge for 16 years and the only thing that helped is having that control (Sophie).

Emma explained that losing control increased anxiety about her child's health: "The thought of not being there to help and check on her, like checking if she's going blue, that's really hard". This seemed to replicate the uncertainty that permeated participants' early parenthood, when their child's survival was unclear, generating hesitance to let go.

Some participants reported further ambivalence from personal losses. Clare, whose child moved to university, described losing her purpose: "I didn't have anywhere near as much to do, 'cause she wasn't there needing my help". For Sophie, this translated into apathy about her wider identity: "As she's becoming more independent, I'm losing that sense of purpose and realising there's nothing else to me". This illustrates that parenting a child with SVCHD can be all-consuming, as letting go not only created practical changes to mothers' daily responsibilities, but also distress at wondering who they were without these roles.

Despite shared reluctance, several participants accepted letting go as necessary and managed the emotional entailments in various ways. Some diligently educated their children

about SVCHD to feel reassured about their ability to cope: "All the things I've learned I've tried to share with him, so that he knows what to do" (Jen). Others tried to prioritise the parent-child relationship. For instance, Clare described reducing contact with professionals following arguments with her child: "We've had a few tiffs lately about me taking over when she speaks to the doctor. They phoned the other day… there was so much I wanted to say, but I had to wait". Whilst mothers used differing coping strategies, there appeared to be a shared feeling that that letting go required emotional resilience.

Conversely, participants whose children had additional needs tried to retain their caring roles. Kerry's child suffered brain damage following multiple cardiac arrests. She stated: "The usual letting go you do as a parent just doesn't happen". These mothers experienced increased fear regarding their child's vulnerability: "I'll go out and walk the same path but the opposite way so I can intercept her in the middle. I'm not great with her independence, the 'what ifs?' are too big a risk for me" (Sophie). Nonetheless, the severe yet chronic nature of SVCHD seemed to make the process of letting go harder for all participants. Indeed, some suggested it is never fully complete because SVCHD carries a reduced life expectancy: "Your role and your worry never stops. You'll never stop trying to look after them" (Jen). There was a sense it was important to maintain connection to caring roles, so that participants would feel prepared to resume these: "I'll be in that carer role again someday… You need to be prepared for that full lifespan, not just until they're 18" (Elise).

In summary, mothers recognised a need to let go of the caring roles they had for their child as they grew older. This process involved anxiety, uncertainty and loss of control beyond that in typical parenthood, due to concerns about their child's health and their own identity without such caring roles. The process of letting go was further complicated by recognition that mothers would need to resume the carer role again someday.

## Discussion

The study addressed the gap in literature by exploring experiences of parental role and identity among mothers of children with SVCHD. Four themes were identified: 1) being a "heart mum", 2) managing competing roles: "you have to wear lots of different hats all at the same time", 3) loss and regaining of identity, and 4) relinquishing control and letting go of caring roles.

The first theme details novel findings regarding the development of a "heart mum" identity, which was informed by disrupted expectations of parenthood and associated guilt and self-criticism. This experience is similar to that described among parents of children with cancer in McKenzie and Curle's (2012) 'End of Treatment Model', which suggests parents are unprepared for changes to their role and identity caused by their child's diagnosis and treatment. The model described a process of 'getting through' treatment, followed by a new period of adjustment. However, there is no end of treatment in SVCHD, which may explain why most participants in this study described a lengthier process of adjustment and lifelong connection to the "heart mum" identity.

Some participants coped with the "heart mum" identity, and associated isolation, by seeking meaning in their difference. However, most tried to avoid it. Similar coping strategies of positive thinking and avoidance are reported among parents of children with various CHD types (Lumsden et al., 2019) and in the initial adjustment phase of the 'End of Treatment Model' (McKenzie & Curle, 2012). However, participants reported not only avoiding negative thoughts but actively trying to hide the "heart mum" identity. This may relate to the concept of perceived parental efficacy, which represents parents' beliefs about their parenting abilities (de Montigny & Lacharité, 2005). Low perceived parental efficacy is associated with low mood, so hiding the identity may have protected mothers from the emotional entailments of feeling unable to meet traditional expectations of parenthood.

Whilst such avoidance may not be adaptive in the long-term, the 'Family Resilience Model' (McCubbin & McCubbin, 1993) suggests that chronic illness involves numerous hardships that can impede immediate adjustment. Instead, it proposes that these families are likely to expeirence a period of maladjustment, because adaptation to the illness evolves over time. Indeed, as their children grew older, participants began to take pride in, and embrace, their "heart mum" identity.

The second theme depicts mothers' struggle to manage additional, competing roles that created fear and exhaustion. The roles were similar to those described in previous research, including protector (Lumsden et al., 2020; Rempel & Harrison, 2007), promoting normality (Lumsden et al., 2020) and a dilemma between being mother and nurse (Elliott et al., 2021; Gaskin, 2018; Harvey et al., 2013), suggesting similarities with other CHD types. This study was the first to describe all four roles together and to discuss the dilemma between normality and protecting the child. Furthermore, the present study extends previous descriptions of protector roles from primarily involving hypervigilance (Lumsden et al., 2020; Rempel & Harrison, 2007) to include advocacy. Fisk et al. (2022) similarly identified advocacy roles as key for parents of children with various CHD types admitted to intensive care following surgery. It may be that the specific nature of SVCHD (i.e., severe yet hidden) increases the necessity of advocacy outside acute hospital environments.

The roles discussed seemed specific to mothers, consistent with gender stereotypes in parenting roles (Katz-Wise et al., 2010). Indeed, mothers discussed frustration that fathers did not have to manage these. Conversely, fathers' accounts in previous research suggest a desire to be more involved (Gower et al., 2017). Interestingly, results extended existing knowledge to include how mothers manage these competing roles, using control, extensive planning, emotional resilience, and mothering through it all. The latter was described by Harvey et al. (2013) among mothers of children with various CHD types during hospital admission for

surgery. This suggests the distress caused by home medical care for SVCHD requires similar coping strategies to those used in hospital, providing insight into the pressure these mothers feel under. Nonetheless, over time, participants became practised in nursing roles and felt better able to balance being 'mum'. This mirrors the process of adaptation through problem-solving and coping in the 'Family Resilience Model' (McCubbin & McCubbin, 1993).

The third theme concerned mothers' experiences of losing and regaining their sense of identity. This finding had not been previously reported, although Gaskin (2018) found parents felt disconnected from their life outside their child's SVCHD. There were clear links with themes one and two, as mothers described feeling consumed by the "heart mum" identity and competing roles, leading to despondence. Similar findings are reported among parents of children with other conditions (Young et al., 2002). Identity theory suggests that parent identities are usually prioritised (Cast, 2004), and a child's vulnerability may present an intensified need for this, as exemplified by participants' descriptions of accepting identity loss in favour of keeping their child safe.

Participants regained their identity over time and this process seemed uniquely impacted by SVCHD, because the risks of the condition created fear of fulfilling identities outside of "heart mum". However, some mothers employed deliberate strategies to avoid identity loss, supporting Pertriwskyj et al.'s (2016) suggestion that parents respond differently to feeling overwhelmed by caregiving identities. In their research with parents of children with disabilities, the authors posit that separation of oneself from the carer identity could be influenced by the child's level of caring need. Similarly, in the present study, Lisa found it easier to maintain other aspects of her identity because her child was unusually healthy and required less medical care. Thus, the acute nature of SVCHD may create differences in parental identity and wellbeing compared to less serious forms of CHD.

The final theme detailed the process and emotional entailments of letting go of caring roles, developing previous descriptions of parental concerns about their child's independence (Lumsden et al., 2020) and transition from paediatric to adult cardiac services (Bratt et al., 2017). Parents of healthy children experience the developmental stage of 'emerging adulthood' as anxiety-provoking and can feel reluctant to promote independence (Kloep & Hendry, 2010). These difficulties are heightened among parents of children with chronic illnesses due to concerns about their child's wellbeing when they lose control over their condition management (Heath et al., 2017). Participants in the current study discussed similar fears. However, they reported more intense ramifications of letting go of caring roles, such as lacking purpose or identity outside of these, highlighting how SVCHD can overwhelm mothers' lives in ways that other conditions may not.

Considering the findings together, there are clear synergies between themes. Being a "heart mum" is informed by various additional, competing parental roles. This impacts mothers' sense of identity and readiness to let go of caring roles. The 'Parenting Under Pressure Model' (Rempel et al., 2013) suggests parents of children with CHD iteratively experience four phases throughout parenthood: 1) encounter a new challenge, 2) adjust, 3) bond with the child, and 4) monitor and protect safety. The findings regarding mothers' experiences and management of changes to their parental role and identity may map onto this model, because challenges, methods of adjustment and efforts to be close to, and protect, their child were evident throughout. The iterative nature of this model also reflects the complex and changing nature of mothers' identity and roles when parenting a child with SVCHD, highlighting how navigating these may differ from conditions with more linear trajectories.

**Clinical implications**

Participants reported difficulties with feeling patronised or judged as 'overprotective' by professionals when advocating for their child, negatively impacting their wellbeing. Therefore, clinical psychologists in multi-disciplinary cardiac teams could provide consultation or training to other professionals (Mercer et al., 2015), including information sharing or collaborative formulation, to support their understanding of the psychological entailments of parenting a child with CHD. This could increase compassion towards parents, allowing them to feel heard and, thus, more comfortable to seek reassurance or medical advice that may improve their child's outcomes.

Many parents of children with CHD manage their child's medical care pragmatically (Lumsden et al., 2019). This may be an adaptive coping strategy, but could prevent support seeking, as in the current study. Therefore, these parents may benefit from support to reflect on their experiences and emotional wellbeing (Gramszlo et al., 2020). Participants reported feeling unable to seek support from existing relationships due to feeling different from parents of healthy children. Thus, health services could facilitate supportive networks between parents of children with SVCHD by establishing group interventions, or co-ordinating with existing charity support networks. Group interventions could normalise parents' experiences and reduce the isolation that participants in this study reported. To do so, they should be consistent, accessible (e.g., online access) and prioritise validation of parents' experiences.

Mothers' experiences of competing roles when parenting their child with SVCHD should be carefully considered given the significant impacts on their identity and psychological wellbeing identified. Research suggests that professionals can underestimate the complexity of managing clinical care in home environments (Kirk et al., 2005), so additional practical support or education may reduce parents' anxiety about nursing roles.

Parents may also benefit from opportunities to discuss how they feel about providing medical care and the impact on their identity, which could be offered through formulation and intervention by a clinical psychologist (Mercer et al., 2015). Access to such clinical psychology support should be available throughout the child's life, given the chronic nature of SVCHD and associated responsibilities.

Additionally, other health professionals could support parents to place boundaries on nursing roles, to reduce the impact of these upon their identity (Woodgate et al., 2015). This could include setting aside time for activities (e.g., reading), as described in the present study. According to Spiers and Beresford (2017), offering professional support with nursing roles (e.g., community nurses) can also allow parents time to be parents. However, the current study found that mothers valued having control over their child's medical care to manage fears about their child's wellbeing, which arose from traumatic experiences of almost losing their child. Therefore, options for professional support should be explored in a non-judgemental and sensitive manner, by recognising that it is understandable to seek control and that accepting support may be challenging. These conversations could be guided by a trauma-informed approach, which seeks to understand the impact of trauma and use this to inform ongoing interactions, to prevent re-traumatisation (Substance Abuse and Mental Health Services Administration, 2014).

As in previous research (e.g., Lumsden et al., 2020), the study highlights the need for targeted support at transitions when parental roles or identity may be acutely challenged. In particular, parents may benefit from support with letting go of caring roles when their child transitions into adulthood. Research shows it is important for parents to understand what to expect during this transition (Bratt et al., 2017), so psychoeducation jointly facilitated by medical teams and clinical or health psychologists may be helpful.

**Strengths and limitations**

The study adds to existing literature by exploring identity and parental role among mothers of children with SVCHD. Prior research had not considered these concepts in depth, potentially overlooking their significance in shaping how parents cope with their child's condition.

Participants were recruited via social media and charity newsletters, so were self-selecting and may have held specific characteristics, such as being emotionally ready to share experiences. The recruitment method could also have excluded those who had not connected with charities or had no internet access, and experiences among these individuals may have differed.

Fathers were not represented in the sample, a limitation across CHD research (Gower et al., 2017), so results may not reflect their experiences. Indeed, mothers noted differing experiences from fathers. For instance, Jen felt her husband gained relief from his 'worker' identity, whilst she lost this. Furthermore, most participants' children were teenagers or adults. Whilst this may have impacted their memories, all participants provided vivid accounts. Nonetheless, interviewing parents still in the medically acute phases of SVCHD may have revealed differences. For example, Holly was the only participant who reported not regaining her identity.

The sample was recruited from the UK, so findings may differ from research in other countries. For example, the financial costs of medical care for CHD in some countries significantly affects parents' experiences (Connor et al., 2010) and might impact perceptions of parental roles. The findings also lack transferability, as they were solely influenced by white, Western perspectives. This is important because CHD is more prevalent in the Middle East (Zimmerman et al., 2020) and research suggests that experiences can differ between Eastern and Western cultures. For example, Im et al. (2018) found the traditional concept of

*TaeKyo*, which suggests interaction with the foetus supports its development, facilitated Korean mothers' acceptance of additional responsibilities (e.g., extra precautions) towards their foetus with CHD.

**Future research**

The study offers novel findings regarding mothers' experiences of identity when parenting a child with SVCHD, particularly the development of a "heart mum" identity. Future research could offer further insights, for example, whether mothers of children with other forms of CHD develop this identity. Grounded theory could also be used to construct theoretical understanding of how the identity develops, because the methodology focuses on generating theory about social or psychological processes and has been used to understand the development of other identities, such as that of 'professional' (Moss et al., 2014).

There is little research into support for parents when their child's health condition brings changes to their role and identity. It would be useful to identify and evaluate possible interventions, using qualitative and quantitative methods, among parents of children with SVCHD, to allow health professionals to offer appropriate support. Mothers in the current study described being unprepared for such changes, leading to guilt, self-criticism and exhaustion trying to balance multiple roles. Thus, psychoeducation regarding changes to parental role and identity, and understandable psychological responses, may be helpful. Ay Kaatsiz & Öz (2020) explored the impact of psychoeducation among mothers of children with cancer and found no change to expectations of parental roles, but some reduction in associated psychological difficulties.

Mothers described that their parental roles and identity were influenced by experiences, such as social interactions, which may also affect children with CHD.

Erikson's model of development suggests adolescence is a critical time for identity formation (Erikson, 1968), but there has been little recent exploration of identity among adolescents with CHD (Luyckx et al., 2011; Rassart et al., 2012). Thus, future research could further investigate this and any associated psychological consequences.

As the study did not include perspectives from fathers, or from individuals outside of white, Western cultures, future research should seek to explore experiences of parental role and identity among these populations to identify similarities and differences, which could inform professionals' ability to tailor support to each parent individually.

## Conclusion

The study explored experiences of parental role and identity among mothers of children with SVCHD. The findings demonstrate that the entailments of SVCHD can significantly impact mothers' parental role and identity and, in turn, their psychological wellbeing. In particular, mothers struggled to balance multiple additional roles and maintain a sense of personal identity. The influence of other factors on these experiences, including the child's health status and interactions with others, cannot be understated. The findings carry implications for how health professionals support mothers of children with SVCHD.

## Data Availability Statement

The data are not publicly available due to privacy and ethical restrictions. Access to supporting data will be granted on a case-by-case basis by the Lancaster University Faculty of Health and Medicine.

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**Table 2.1**

*Participant demographic data*

| Pseudonym | Child's Age at Interview | Child's Position in Family | Child's Sex | Child's Condition | Time of Diagnosis | Time since Fontan |
|---|---|---|---|---|---|---|
| Emma | 15 | 1st of 3 | F | Tricuspid Atresia | Prenatal | 10 years |
| Holly | 4 | 3rd of 3 | F | HRHS†, Pulmonary Atresia | Prenatal | 9 months |
| Kerry | 22 | 1st of 3 | F | Hypoplastic Left Heart Syndrome | Prenatal | 18 years |
| Clare | 19 | 1st of 3 | F | Pulmonary Atresia | Postnatal | 13 years |
| Jen | 19 | 1st of 3 | M | HRHS, Pulmonary Stenosis, TGA‡ | Postnatal | 15 years |
| Sophie | 16 | 1st of 2 | F | HRHS, CAVSD§ | Postnatal | 11 years |
| Elise | 20 | 1st of 2 | M | Tricuspid Atresia, TGA | Prenatal | 16 years |
| Lisa | 18 | 1st of 3 | M | Hypoplastic Left Heart Syndrome | Prenatal | 15 years |

† Hypoplastic Right Heart Syndrome
‡ Transposition of the Great Arteries
§ Complete Atrioventricular Septal Defect

**Figure 2.1**

*Conceptual model of themes*

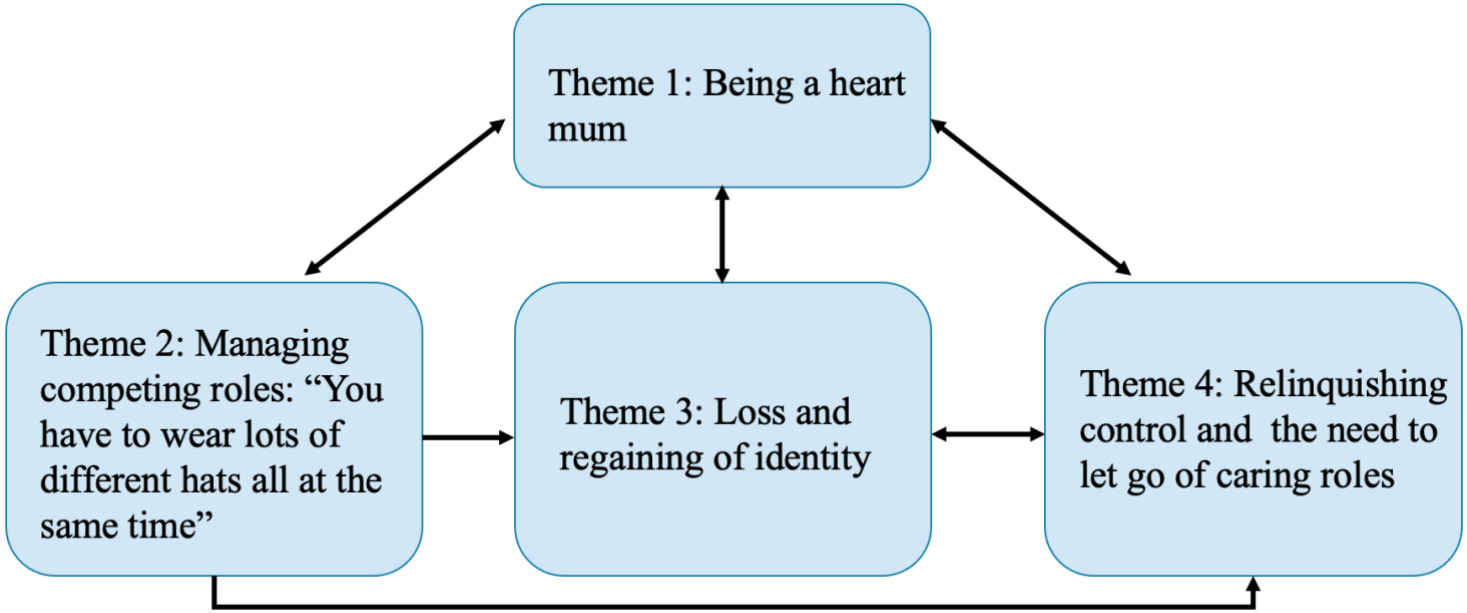
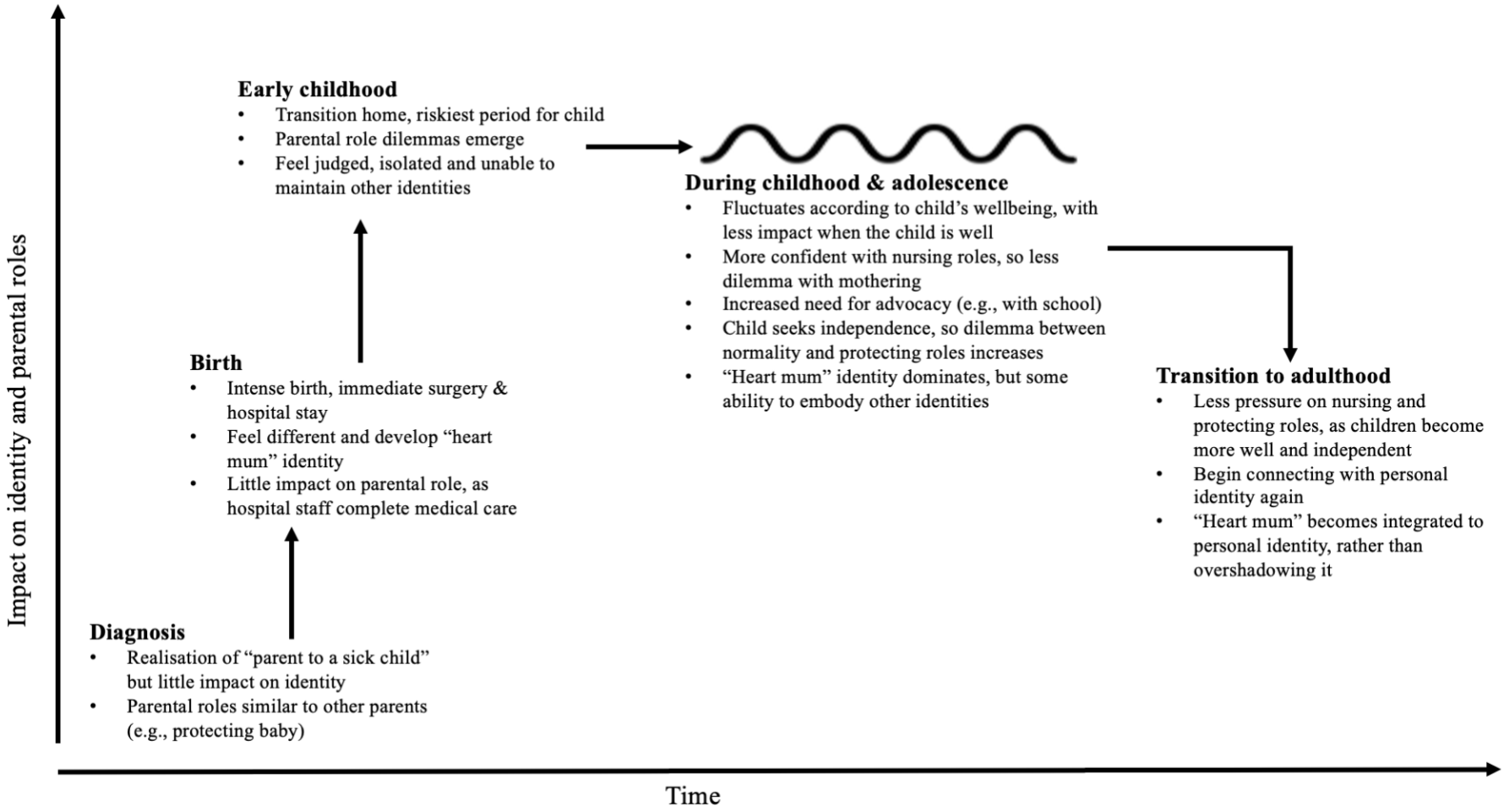


**Figure 2.2**

*Participants' experiences of parental role and identity over time*



## Appendix 2-A
## Statement of Contribution

*What is known on this subject?*

- Parents of children with congenital heart disease (CHD) are more likely to experience mental health difficulties than parents of healthy children.
- Parents' perceptions of their parental role and identity can affect their psychological wellbeing and are critically impacted when their child has a health condition.
- Research among parents of children with CHD suggests mothers experience difficulty balancing mothering and nursing roles.

*What does this study add?*

- Mothers described a "heart mum" identity, which was reported to consume their personal identity.
- Balancing parental roles involved challenging emotional and practical responsibilities.
- Mothers found it difficult to let go of caring roles, with implications for their coping.

## Appendix 2-B
## Murray and Wilde (2020) IPA Methodology

1. Choose a transcript to begin with and format it for analysis.
2. Read the transcript with the research questions in mind and make notations (initial coding).
3. Copy notations to Post-it notes.
4. Group Post-it notes into clusters (grouping codes).
5. Write extended narratives for each cluster.
6. Produce a table for each cluster, with original notations, narratives, and supporting quotes.
7. Title the cluster.
8. Repeat steps 1-7 for subsequent transcripts.
9. Merge analysis across transcripts.

**Appendix 2-C**
**Example of coding**

| Initial Codes | Transcription |
|---|---|
| Being the "parent of a heart child" involves lots of roles at once.<br><br>"Bad cop" saying 'no' due to the condition vs. pushy to have normality and independence.<br><br>Supporting the child to live a normal life filled with experiences.<br><br>As the child grows older there is a new role to support her to take control of her health.<br><br>Mum describes letting go of caring roles as 'weird' – a big change.<br><br>Anxiety about not being able to check on her child and losing control over caring roles.<br><br>Losing control over caring roles creates uncertainty about wellbeing.<br><br>Having to prepare her for the world more than other children.<br><br>Preparing her child by sharing information and practising medical care together to provide reassurance that she knows what to do. | Your role changes a lot and again that's probably the same for any parent but being the parent of a heart child, you have to wear lots of different hats all at the same time: gobby mum, ambassador, nurturer, the bad cop. That's when there's a new activity or something they want to do and you say no because of their condition, but also when they say they can't do something that you know they can. I've been more forthright and pushy with her than with my other children because I want her to recognise all the things she can do and for other people to recognise that too. I want to support her to try new stuff and live as normal a life as possible. Now we're in the process of getting her medical records because she'll be 16 in summer and wants to go to uni in a few years and she knows what she wants to do. But my role now is to support her to take control of her health, it's been weird trying to let go. Whilst I want her to be independent, the thought of her not being here and me not being there to help her and check on her, like checking if she's going blue, that's really hard *tearful*. The medical stuff has been okay since she was younger and it's more the psychological stuff we've been dealing with recently. She wants to go into medicine though so now we're trying to help support her with her understanding her own condition and she can tell people and know how to ask for help. We have to prepare her for the world much more than other children because she's at such a risk, like her lungs don't work well and especially with COVID-19 that's important. She also has asthma, which is heart generated, so I need to make sure she knows and is prepared to look after herself. So now we do things together like her INRs and warfarin (finger pricking), she doesn't like it, but I make her do it but while she's still here we need to start because the next few years will fly by. |

**Appendix 2-D**
**Theme from participant one's audit trail**

*"Gobby mum": Protecting and advocating for the child*

| Initial Codes | Narrative Summary | Illustrative Quotes (Page:Line) |
|---|---|---|
| • Mum describes locking emotions away to protect the child<br>• Quickly learning to be a "gobby mum" to assert expertise in her own child and their needs<br>• Mum felt concerned about being viewed as a "pain in the backside"<br>• Being right and listened to led to realisation of expert role and confidence to advocate<br>• Challenging medical professionals is uncomfortable but necessary<br>• Anger at being seen as "pushy" or "anxious" mum<br>• Hidden nature of the condition requires mum to be an advocate but means she is viewed as 'over-protective'<br>• Viewed as "assertive parents" and admired for challenging professionals | This theme reflects Emma's efforts to protect and advocate for her child. On a personal level, Emma believed that managing and hiding her emotions was one way to protect her child. There was a sense of not wanting to add to her child's struggles given everything she had been through with her physical health. Emma also talked of wanting to ensure positivity in the relationship, particularly in her child's early years.<br>Primarily, in protecting and advocating for her child, Emma described herself as a "gobby mum", both internally and in how others perceived her. This was not something she had previously identified with, and she found it uncomfortable at times. This indicates that Emma was willing to sacrifice her authentic self to ensure her child's needs were met, even at the expense of others' perceptions of her. There was a suggestion that Emma felt frustrated and somewhat embarrassed by having to be "gobby". However, she rationalised the need for this role following experiences of not being listened to and something bad then happening to her child. This suggests that the role was driven by underlying fear and concern that permeated her experience of being a parent to a child with SVCHD. | • "I learned quickly that I need to not even just be assertive but to be a 'gobby mum', in a different way to being chatty or positive but in a way of standing up for your child and asserting your expertise in knowing your child and saying, 'this isn't right'" (9:9)<br>• "It's amazing how that turned my role around as a parent because I suddenly realised "I knew that" and became more of an expert in my child and more of an ambassador for them" (9:20)<br>• "We had to really fight to get mental health support for her because there really isn't any in our area. That whole 'gobby mum' and 'pushy mum' had to come out and annoy my GP, everyone, until someone gave us some support" (10:2)<br>• "Within a week I'd been back to the GP 3 times and being that 'pushy mum' and was really patronised, like patting me on the head saying, 'it's fine mum, it's your first born don't worry'" (14:16)<br>• "I guess, I find it frustrating but it's a learning curve that you need to have a different role for a heart child, you need to push against medical |

| | | |
|---|---|---|
| | Although protecting and advocating for her child made sense to Emma as it facilitated her child's needs being met, she did express concern about how others perceived her. Whilst some parents had admired assertiveness with professionals, Emma seemed to feel embarrassed when recognising that other parents would probably respond differently. Indeed, she discussed how medical professionals could be patronising, because they viewed her as over-protective, which created anger. Ultimately, Emma seemed to experience loneliness and isolation in this role, as she found it hard to identify with other parents and professionals. | professionals and make a fuss, which can be uncomfortable" (14:24)<br>• "I think lots of parents find that hard because these are professionals who should be listened to, and I do still believe that, but I also really have a much greater role in disagreeing with them too and knowing that I have the right to do something about that" (14:29)<br>• "She needed me to advocate more because often her condition was hidden, so there'd be things she couldn't do but people wouldn't understand why" (22:19)<br>• "We become those assertive parents, especially with professionals, and people see that as quite ballsy and not something they would ever do. They seem to think it's quite cool" (23:10) |

**Appendix 2-E**
**Participant contributions to final themes**

| | **Being a "heart mum"** | **Managing competing roles: "You have to wear lots of different hats all at the same time"** | **Loss and regaining of identity** | **Relinquishing control and letting go of caring roles** |
|---|---|---|---|---|
| Emma | • What it means to be parent to a child with SVCHD. | • Striving for normality and positivity.<br>• Navigating medical care.<br>• "Gobby mum": Protecting and advocating for the child. | • Loss and reappraisal of identity. | • Preparing the child for the world and struggling to "let go". |
| Holly | • "Heart mum" as different to any other parental identity.<br>• The role of others in making sense of identity. | • One step ahead: Facilitating normality through preparation.<br>• Balancing being 'mum' and 'nurse'.<br>• "Mamma bear": the role of protection. | • "I can't be the same person I was before": Loss of identity. | |
| Kerry | • The reality of parenting a 'sick' child. | • Mothering through it all.<br>• "I can never just be mum": The need for a nursing role.<br>• Balancing normality with protecting her child. | • Renegotiating a of sense of identity. | • Balancing normality with protecting her child. |
| Clare | • Renegotiating identity: What it means to be "heart mum". | • Managing competing identities of 'carer' and 'mum'.<br>• Seeking protection vs. normality for the child. | • Renegotiating identity: What it means to be "heart mum". | • "It's like a rollercoaster": Learning to let go. |

| | | | | |
|---|---|---|---|---|
| Jen | • What it means to be 'mum' in the context of SVCHD. | • What it means to be 'mum' in the context of SVCHD.<br>• Supporting the child to live life to the fullest: Roles of encouragement and protection | • Feeling powerless to be herself: Loss of identity and appraisal by others.<br>• Changes with age: Letting go of parental roles and reconnecting with identity. | • Changes with age: Letting go of parental roles and reconnecting with identity. |
| Sophie | • Being the "parent of a disabled child" | • Being on "autopilot": Trying to balance being 'carer' and 'mum'.<br>• Appreciating and making the most of time with her child: The importance of normality.<br>• Protecting her child regardless of the consequences for herself. | • "It becomes all you are": Loss and efforts to regain identity. | • The struggles of letting go. |
| Elise | • Identity changes: The struggle to retain a sense of self after becoming "parent to a disabled child". | • The struggle of striving for normality.<br>• A role for mum: Becoming a carer. | • Identity changes: The struggle to retain a sense of self after becoming "parent to a disabled child" | • A role for mum: Becoming a carer. |
| Lisa | • Being parent to a disabled child: Feeling "robbed" of normal parenthood. | • Making the most of life and striving for normality.<br>• A carer role influenced by medical circumstances and need for independence.<br>• A constant battle: The role of protection. | • Loss of identity: Feelings of failure and resentment. | • A carer role influenced by medical circumstances and need for independence. |

# Section 3: Critical Appraisal

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# Critical Appraisal

The aim of this critical appraisal is to summarise the findings from the systematic literature review and empirical paper, and to provide critical reflections on the research. There will be discussion of methodological considerations, reflexivity, limitations and implications of the research, as well as personal reflections on research processes.

## Selecting the Research Topic

The decision to focus the research on parents' experiences of having a child with congenital heart disease (CHD) was influenced by my experience in clinical health psychology settings. In particular, I worked for two years as an assistant psychologist in a cardiology team at a northwest tertiary children's hospital. I became aware of the additional challenges that children with CHD encounter, including hospital admissions and feeling different (Chong et al., 2018), and developed a passion for supporting them and their families. Although my clinical work was mainly with children, I also noticed the psychological impact of CHD on their parents (Biber et al., 2019). Indeed, guidance at the time emphasised the importance of psychological support for parents at any stage in their child's care (NHS England, 2016). However, in practice, I observed an understandable prioritisation of the child's needs, meaning those of parents could be overlooked by professionals. Furthermore, in meetings with clinical psychologists working in cardiology teams across the UK, there were often discussions regarding the need for further research with parents. Thus, I was aware that research in this area would both address gaps in the literature and have the potential to influence clinical practice.

Whilst I felt my passion for, and knowledge of, the area was advantageous, I was mindful it could unintentionally influence the research process. For instance, and of particular relevant to Interpretative Phenomenological Analysis (IPA), attachment to the research topic

can increase the risk that preconceptions unduly influence the research (Smith et al., 2009). From the outset, I sought to manage this through regular reflection in supervision, thorough documentation of audit trails and maintaining a reflective diary.

## Section 1: Systematic Literature Review

### *Summary of findings*

The systematic literature review synthesised qualitative research regarding parents' experiences of receiving their child's CHD diagnosis. Using thematic synthesis, as outlined by Thomas and Harden (2008), four main themes were identified, although experiences of the emotions discussed varied between individuals. There was consistently unpreparedness to receive the diagnosis, creating shock, guilt and anger. Parents then faced a period of overwhelming information, decision-making and uncertainty about their child's prognosis. Parents' initial experiences were especially influenced by interactions with professionals, with clear information and compassion valued. As parents realised the reality of CHD, they mourned the loss of 'normal' pregnancy or birth experiences, and of their envisioned healthy child. However, they utilised various coping strategies throughout to reach a position of accepting the diagnosis, which involved adjusting their expectations of pregnancy, birth and their future.

### *Methodological considerations*

The research question was informed by the thesis preparation assignment, completed in March 2020 during my first year of training. The assignment found several studies had explored parents' experiences of their child's CHD diagnosis, but no synthesis of these findings had been published. I sought to address this gap, because systematic literature reviews provide comprehensive overviews of, and identify gaps in, existing research

(Flemming & Noyes, 2021). I chose to synthesis qualitative evidence, because qualitative methodology was consistent with my approach to the empirical paper and can explore detail regarding parents' experiences that quantitative synthesis cannot.

When planning the synthesis, I intended to use meta-ethnography, because it seeks to synthesis findings into a whole that generates new understandings (Noblit & Hare, 1988), which corresponded with the aim to explore parents' experiences. However, after completing the systematic search and data extraction, I realised there was a mixture of descriptively 'thin' and 'rich' data. Meta-ethnography requires conceptually 'rich' data to enable further interpretation, so was not an appropriate methodology (France et al., 2019). Following discussions with the research team and examination of guidance for qualitative evidence synthesis, I chose to use thematic synthesis. Thematic synthesis can be used to synthesis both 'thin' and 'rich' data, whilst still allowing interpretation (Flemming & Noyes, 2021), and Booth et al. (2016, p.22) argue it is "more epistemolgy-neutral" because it adopts a critical realist approach. Thus, it allows the integration of studies that use various qualitative methods into a single synthesis, which is valueable to inform policy and practice in health services. This was relevant, because the review was likely to consider parents' support needs following their child's diagnosis and how health services could meet these.

The Critical Appraisal Skills Programme Qualitative Checklist (2018) was used to assess the quality of included studies because it provides a clear framework that is accessible to novice researchers, like myself, and has been deemed suitable in guidance for qualitative evidence synthesis (Long et al., 2020). However, its checklist nature may mean that quality appraisal becomes focused on the paper's reporting of the methodology, rather than the value of the research (Majid & Vanstone, 2018). Alternatively, Walsh and Downe's (2006) checklist is less prescriptive and may have promoted more consideration of the contribution

of the research as a whole. Nonetheless, Majid and Vanstone (2018) argue the less prescriptive nature can create difficulties with systematically reporting the quality appriasal.

### *Reflections on the findings*

Prior to the thesis, I had little experience in academia. To manage my anxiety about getting things 'wrong' and enhance the credibility of the synthesis, several steps were taken. For example, the publication year of included articles was not limited, two independent reviewers assessed 10% of titles and abstracts and 25% of full texts against eligibility criteria to reduce error or bias in the selection process, one independent reviewer assessed the quality of all included studies and, finally, quotes were provided to evidence all themes.

Nonetheless, there were some limitations, as outlined in section 1. In particular, the criteria for papers to be published in peer-reviewed journals was used because it lent support to the validity of included studies. However, this criterion can generate publication bias and may have excluded other findings. For example, grey literature can add valuable or novel contributions to systematic reviews, although this can be outweighed by the considerable time required to locate and synthesise it (Mahood et al., 2013). Indeed, grey literature identified by the scoping searches comprised conference abstracts with no participant quotes, so I chose not to include this source of information in the systematic search.

The systematic literature review had implications for research and clinical practice. One key finding was the influence of professionals on parents' emotional wellbeing when receiving their child's CHD diagnosis. Whilst some research shows these professionals are at risk of burnout (Cohen et al., 2020), there has been little exploration of their experiences of informing parents of their child's diagnosis, so this should be prioritised in future research. Additionally, a corresponding recommendation for was these professionals to have access to regular supervision or reflective practice facilitated by a clinical psychologist. Future

research could evaluate any implementation of this recommendation to determine its effectiveness.

## Section 2: Empirical Paper

### *Summary of findings*

The empirical research aimed to explore how having a child with single ventricle CHD (SVCHD) affected parents' experiences of parental roles and identity. Eight mothers were interviewed, and data were analysed using IPA (Murray & Wilde, 2020). Four themes were identified: 1) being a "heart mum", 2) managing competing roles: "you have to wear lots of different hats all at the same time", 3) loss and regaining of identity and 4) relinquishing control and letting go of caring roles. The findings illustrate how the acute yet chronic nature of SVCHD can challenge mothers' parental roles and leave them feeling their personal identity is consumed by caring responsibilities. Similar findings have been reported among parents of children with cancer (McKenzie & Curle, 2012). However, some differences were noted; for instance, there was more emphasis on the lifelong nature of the "heart mum" identity and the intensity of distress resulting from parental role dilemmas. There were also differences between participants, related to the child's age and physical health status, which highlights the complex nature of the findings and the need to further explore parental role and identity among parents of children with CHD.

### *Methodological considerations*

**The use of IPA.** IPA was chosen to analyse the data because the underlying principles of phenomenology and idiography, which respectively concern the essential aspects of a phenomena and the uniqueness of each individuals' experience (Pietkiewicz & Smith, 2014), corresponded with the aim to explore experiences of parental role and identity.

I also felt a strong commitment to ensure individual voices were not lost to the analysis, which can occur in other qualitative approaches in which larger samples are used and there is emphasis on commonalities. The third principle underpinning IPA, hermeneutics, concerns the researcher's role in interpreting participants' experiences and requires particular attention to 'bracketing' preconceptions (Smith et al., 2009). I noticed tension between using my prior experience to inform my understanding of the topic area and ensuring it did not unduly influence the analysis. Thus, such careful 'bracketing' of preconceptions prior to each interview and throughout the analysis was important to allow me to privilege participants' accounts.

Moreover, IPA adopts a realist epistemological stance; it recognises there may be multiple 'realities' and assumes that careful use of language and interpretation can provide insight into the experiences of 'reality' for individual participants. This fits with my own epistemological stance of critical realism, whereby I believe that an objective reality exists, but the way we perceive it is subjective and influenced by social context (Cuthbertson et al., 2020).

**Recruitment.** A recruitment strategy via a tertiary children's hospital cardiology team had initially been arranged. However, due to the onset of Covid-19 pandemic in 2020, the service became unable to facilitate recruitment. Consequently, the research was instead advertised via the social media, websites, newsletters and email lists of UK heart charities. The large reach of charities could have compromised the homogeneity required in IPA to produce a detailed analysis of phenomena (Smith et al., 2009). However, SVCHD is a rare condition, accounting for approximately 0.093 per 1,000 live births (Liu et al., 2019), so I felt an appropriate level of homogeneity could be maintained.

Despite the large following of the charities who promoted the research, there were difficulties with recruitment. Therefore, I further amended the recruitment strategy in

February and June 2021 to create social media pages dedicated to the research, which allowed me to advertise in ways that charities could not (e.g., not wanting their page dominated by repeated research advertisements). However, recruitment continued to be slow. I had received contact from several interested individuals who had been ineligible to take part, due to criteria that their child had to be aged 16 years or under. I weighed the possibility of increasing the age range to accommodate these individuals and recruit a sufficient sample against the potential of reducing homogeneity among the sample by doing so. I decided to increase the age limit of participants' children to 25 years in July 2021, to support recruitment, because the NHS Long Term Plan (NHS England, 2019) outlines plans to implement an age range of 0-25 years in children's medical and mental health services. Increasing the age limit of participants' children enabled the research to capture experiences of transition to adulthood, which informed the final theme 'relinquishing control and letting go of caring roles', and otherwise might have been missed.

**Conducting interviews during Covid-19.** Due to the Covid-19 pandemic, interviews had to be conducted online via video-conferencing software. As I could not account for when lockdowns might end or next occur, I decided to continue with remote interviews throughout data collection to maintain consistency. Whilst there are advantages to in-person interviews, research suggests that completing interviews over video-conferencing software does not compromise the quality of data collected (Gray et al., 2020; Krouwel et al., 2019). However, a key concern was that some research does suggest it can be more difficult to build rapport during remote interviews (Roberts et al., 2021). Interviews via video-conferencing software may be less affected than those via telephone, as visual cues are still available to consider how participants might be feeling (e.g., body language) and encourage them to continue (e.g., nodding). Nonetheless, to manage this concern, I allocated additional time to facilitate rapport building and used more verbal cues to demonstrate my interest (e.g., mm or ahh).

Participants appeared to feel comfortable to share their experiences, evidenced as the shortest interview lasted for 72 minutes. Indeed, remote interviews may even have benefitted the research because online formats can be less intimidating and increase openness (Weller, 2017).

Whilst the research took place using the Covid-19 pandemic, the aim was to explore experiences of parental role and identity throughout the child's life and not specific to Covid-19. Therefore, I chose not to directly address the pandemic in the interview schedule. Although some participants did discuss Covid-19, it was not an area of sufficient focus to merit inclusion in the final analysis. These participants stated that the pandemic did not significantly alter their experience of their parental role, rather, that others had become more 'like them', for example, being more protective and cautious about germs.

### *Ethical considerations: Managing distress*

Parents of children with SVCHD are considered vulnerable to experiencing psychological distress (Woolf-King et al., 2017). I was aware that such distress might have been exacerbated by the Covid-19 pandemic; for example, these parents report increased fear due to their child's vulnerability to Covid-19 (Marino et al., 2021). I was mindful that existing distress coupled with the sensitive nature of the research could make it upsetting for participants to share their experiences. I considered this in advance and took steps to minimise potential distress, including reminding participants that they could take a break or withdraw at any time, scheduling time for a verbal debriefing at the end of interviews and offering to share written information containing sources of support via email.

Although no adverse events arose, some participants were tearful during their interviews. I used my clinical skills to respond with active listening and empathy, which allowed individuals to feel understood (Weger et al., 2014). However, my role during

interviews was to listen to participants' experiences, not offer psychological support and, as I felt a pull throughout interviews to summarise, validate and reassure, it generated tension in me between being a clinician and a researcher. I used my reflective diary to consider the impact of this tension on me:

> Struck by the fact that some participants have never spoken about their experiences of parenting a child with SVCHD and feeling privileged that they have chosen to share these with me. Noticing guilt at feeling unable to offer normalisation, support and containment whilst they discuss experiences of such vulnerability (13/08/2021).

I reflected on this further during thesis supervision and considered how to approach future interviews. For example, it was possible to offer some summaries during the interview to confirm my understanding of the information shared. Whilst this did not amount to psychological support, it allowed me to feel reassured that I was not potentially invalidating participants' experiences by sticking solely to the interview topic guide.

***Data analysis***

Although analysis in IPA has several typical features (such as analysing each transcript separately and 'bracketing' analysis between transcripts before merging analysis across transcripts), its principal proponents do not dictate a specific analysis procedure. I chose to follow the approach outlined by Murray and Wilde (2020), because I found it to be an accessible guide and it was co-written by my thesis supervisor, so I was able to draw upon their expertise throughout. The approach, for example, includes a number of processes designed to develop the interpretative aspect of IPA that many novice researchers struggle to achieve. It also prioritises capturing patterns and divergences in one narrative, rather than presenting several subthemes for each theme. Although I initially found this challenging to produce, it supported my desire to prioritise participants' voices through nuanced narratives.

One aspect of the approach that facilitated this, which differs from some guides to IPA, was writing and titling interpretative summaries for each theme for each participant, before analysing the next transcript. Whilst writing the summaries was time-consuming, it increased my familiarity with the data and encouraged interpretation, rather than description, of participants' experiences. It also produced a thorough audit trail, lending credibility to the analysis.

### *Reflexivity*

Reflexivity involves self-reflection by the researcher on their potential biases and relationship to the topic being explored, to understand how they may influence the research (Berger, 2013). Reflexivity is particularly important in IPA, because the underlying principle of hermeneutics recognises that researchers' interpretations of participants' accounts are affected by their own assumptions. Thus, a fundamental process in IPA is the 'bracketing' of preconceptions to prevent undue influence on the research (Smith et al., 2009). To manage this, I kept a reflective diary to document my thoughts and any identified assumptions, and regularly discussed these in supervision. For example, following the first interview, I felt surprised by the participant's description of adaptive coping to changes in their parental role and identity, and positive impacts of SVCHD on their parenting experience:

> Feeling surprised by the optimism shown in such overwhelmingly difficult circumstances and by the positives identified along the participants' journey, as well as their navigation of these situations without accessing support (16/04/2021).

Whilst I understood that parents would develop coping strategies to manage their child's CHD, and I had become familiar with previous research evidencing this (Lumsden et al., 2019), my assumptions may have been influenced by prior clinical experiences. In my role as an assistant psychologist, I supported children with CHD and their families who were

experiencing difficulties. Thus, the dominant narratives about parenting a child with SVCHD were negative, concerning difficulties with coping and the need for support. This could have dictated my line of questioning or limited my exploration of coping during interviews. However, explicitly recognising and 'bracketing' these preconceptions allowed me to prioritise participants' accounts and recognise important aspects of coping with alterations to their parental role, for example, navigating dilemmas between mothering and nursing roles.

### *Reflections on the findings*

I tried to enhance the credibility of the findings by promoting homogeneity using a UK sample, ensuring themes were evidenced by at least half of the participants, as recommended (Smith, 2011), and keeping thorough audit trails. However, there are study limitations. In particular, following completion of the research, I reflected on some perspectives that might not have been represented.

Fathers were not included in the sample, which is a limitation across the research area (Lin et al., 2021). During recruitment, no fathers expressed interest in the research. In quantitative research by Davison et al. (2017), fathers indicated they were less likely to participate in paediatric research because they were too busy, not encouraged by mothers or had simply not been asked. On reflection, I could have specifically directed some study advertisements towards fathers after noticing that only mothers were being recruited. Nonetheless, research with fathers of children with CHD shows that they often believed their input to their child's care was not as valued as mothers' and, subsequently, felt excluded from parental roles (Hoffman et al., 2021). Thus, the research topic may not have resonated with fathers. Whilst mothers shared some views regarding fathers' parental role and identity (e.g., less impact on fathers' identity because mothers adopted most caring roles), research should seek to explore fathers' perspectives directly, as these may differ. Indeed, the systematic

literature review illustrates that fathers often hide their true feelings from their partners to fulfil gendered social roles of being 'strong'.

Additionally, all but one participants' child with SVCHD was their first-born. It is important to consider how this may have impacted their parenting of subsequent children. For example, Clare discussed that SVCHD created a general fear of harm that made her "more cautious" with all of her children. Thus, parents whose first-born child has SVCHD may be less attuned to how the condition impacted their parental roles and identity compared to those who experienced parenting a healthy child first. Future research should recruit samples with more diversity in the birth order of children to better explore the specific impacts of SVCHD on parental role and identity.

**Personal Reflections**

The process of completing the thesis was both challenging and rewarding, and it has been important to consider the impact on me as both a researcher and clinician. Prior to training, I doubted my abilities to successfully design and lead a thesis research project and felt daunted at the prospect of doing so. Although the research has been demanding and, at times, frustrating, it has provided opportunities to challenge these assumptions about myself. Indeed, developing my research skills, producing impactful findings with implications for clinical practice and interacting with participants has motivated me to continue my involvement in research post-qualification. Clinician involvement in health research is important, as it can promote the study of 'real-world issues' that matter to individuals who access services (Yanos & Ziedonis, 2006).

As a clinician, I have a longstanding passion for clinical health psychology and have secured a role as a clinical psychologist in a paediatric clinical health psychology service. The thesis research reaffirmed my commitment to supporting parents, as well as children, in

these services and to work with the multi-disciplinary team as a whole to support parental wellbeing. For instance, I was struck by participants' descriptions of difficult interactions with professionals, during which they felt dismissed or patronised when trying to advocate for their child. This experience has enhanced my passion for person-centred care and prioritising the voice of the individual and their family. Indeed, a key role for clinical psychologists in health settings is to offer consultation and training to multi-disciplinary teams to support their understanding of the psychological impact of medical conditions on patients and their families (British Psychological Society [BPS], 2008; Mercer et al., 2015).

**Dissemination plan**

A summary of the empirical paper was presented to first and second-year trainee clinical psychologists, and research staff, as part of the Lancaster Doctorate in Clinical Psychology, at the thesis presentation day in April 2022. Both the systematic literature review and the empirical paper will be submitted for publication in the British Journal of Health Psychology by the end of August 2022. As one of the thesis supervisors is a chartered member of the BPS, it will be possible for the research to be published as open access. Open access publication will promote wider access to the findings and, it is hoped, enable more services to consider the clinical implications. Finally, a summary of the empirical paper will be sent to all participants who consented to receiving this, and the charities involved in advertising the research, to thank them for their contributions and inform them of the findings. The lay summary will also be published on the Perinatal Mental Health and Parenting Research Unit website, to further enhance the accessibility of the research.

## Conclusion

The thesis explored parents' experiences of receiving their child's diagnosis of CHD and of parental role and identity when parenting a child with SVCHD. The critical appraisal offers insight into the research process and key decisions that were made, including methodological and ethical considerations and reflections on the limitations of both papers. The appraisal highlights the importance of reflexivity throughout and details some specific reflections about the research process and the impact on the researcher.

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# Section 4: Ethics Section

## Ethics application for research paper: Parenting a child with single ventricle congenital heart disease: Mothers' experiences of role and identity

Shannon Dandy

Doctorate in Clinical Psychology

Lancaster University

Word Count: 5875 (Excluding references and appendices)

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

**Application for Ethical Approval for Research**

***for additional advice on completing this form, hover cursor over 'guidance'.***

**Guidance on completing this form is also available as a word document**

**Title of Project**: Having a child with single ventricle congenital heart disease: Parental experience of role and identity

**Name of applicant/researcher**: Shannon Dandy

**ACP ID number (if applicable)*:** N/A **Funding source (if applicable)** N/A

**Grant code (if applicable):** N/A

***If your project has *not* been costed on ACP, you will also need to complete the Governance Checklist [link].**

**Type of study**

☐ Involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants. **Complete sections one, *two* and four of this form**

☒ Includes *direct* involvement by human subjects. **Complete sections one, *three* and four of this form**

**SECTION ONE**

**1. Appointment/position held by applicant and Division within FHM** Trainee Clinical Psychologist, Doctorate in Clinical Psychology

**2. Contact information for applicant**:
**E-mail**: s.dandy@lancaster.ac.uk **Telephone**: 07411135890 (please give a number on which you can be contacted at short notice)

**Address**: Health Innovation Campus, Lancaster University, Bailrigg, Lancaster, LA1 4YG

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

Dr Craig Murray (Academic Supervisor), Senior Lecturer, Division of Health Research, Lancaster University, Lancaster LA1 4YG

Dr Anja Wittkowski (External Academic Supervisor, DClinPsy), Senior Lecturer, Doctorate in Clinical Psychology, University of Manchester, Manchester, M13 9PL

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

PG Diploma ☐ Masters by research ☐ PhD Thesis ☐ PhD Pall. Care ☐

PhD Pub. Health ☐ PhD Org. Health & Well Being ☐ PhD Mental Health ☐ MD ☐

DClinPsy SRP ☐ [if SRP Service Evaluation, please also indicate here: ☐] DClinPsy Thesis ☒

**4. Project supervisor(s), if different from applicant**: Dr Craig Murray

5. **Appointment held by supervisor(s) and institution(s) where based (if applicable)**: Senior Lecturer, Lancaster University

**SECTION TWO**
**Complete this section if your project involves existing documents/data only, or the evaluation of an existing project with no direct contact with human participants**

1. Anticipated project dates (month and year)
Start date: End date:

2. Please state the aims and objectives of the project (no more than 150 words, in lay-person's language):

**Data Management**
*For additional guidance on data management, please go to* Research Data Management *webpage, or email the RDM support email:* rdm@lancaster.ac.uk
3. Please describe briefly the data or records to be studied, or the evaluation to be undertaken.

4a. How will any data or records be obtained?

4b. Will you be gathering data from websites, discussion forums and on-line 'chat-rooms' n o
4c. If yes, where relevant has permission / agreement been secured from the website moderator? n o
4d. If you are only using those sites that are open access and do not require registration, have you made your intentions clear to other site users? n o

4e. If no, please give your reasons

5. What plans are in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.

6a. Is the secondary data you will be using in the public domain? n o
6b. If NO, please indicate the original purpose for which the data was collected, and comment on whether consent was gathered for additional later use of the data.

Please answer the following question *only* if you have not completed a Data Management Plan for an external funder
7a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?

7b. Are there any restrictions on sharing your data?

**8. Confidentiality and Anonymity**
a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications? yes
b. How will the confidentiality and anonymity of participants who provided the original data be maintained?

9. What are the plans for dissemination of findings from the research?

10. What other ethical considerations (if any), not previously noted on this application, do you think there are in the proposed study? How will these issues be addressed?

**SECTION THREE**
**Complete this section if your project includes *direct* involvement by human subjects**

**1. Summary of research protocol in lay terms (indicative maximum length 150 words):**
Congenital heart disease (CHD) means there is problem with the structure of the heart. Children with CHD and their families are at increased risk of experiencing mental health difficulties, especially if the condition is serious (e.g., single ventricle CHD [SVCHD]). Previous research into parents' experiences has recognised impacts upon their perceptions of their own role or identity. For example, some describe difficulty trying to be both parent and nurse to their child. However, no study has examined this in detail. The proposed research seeks to address this gap and will be advertised via UK heart charities. Parents of children with SVCHD will be interviewed about their perceived role, identity and psychological wellbeing. Interviews will be recorded and analysed using Interpretative Phenomenological Analysis (IPA). It is hoped that findings will develop understanding of parents' psychological needs and shape support offered by professionals.

2. **Anticipated project dates (month and year only**)
Start date: February 2021 End date: March 2022

**Data Collection and Management**
*For additional guidance on data management, please go to* Research Data Management *webpage, or email the RDM support email:* rdm@lancaster.ac.uk

**3. Please describe the sample of participants to be studied (including maximum & minimum number, age, gender):**
The research aims to recruit 6-12 participants, as a small, defined sample is recommended when using Interpretative Phenomenological Analysis (IPA) because of its idiographic focus (Murray &

Wilde, 2020). The sample will be based in the United Kingdom. Participants will be parents of children with SVCHD who have had their completed Fontan surgery, which is the last of three stages of surgery in usual treatment. There are specific inclusion and exclusion criteria for the research:

Participants will be eligible to take part if:
- They have a child with SVCHD
- The child had their completed Fontan surgery at least six months prior to the interview
- The child is aged 25 or under
- They reside in the UK
- They are able to comprehend and speak English

*Notes on inclusion*
- The rationale for the child having had the completed Fontan surgery is to allow exploration of the complete surgical journey and promote homogeneity among participants. The requirement for the child to be at least 6 months post-surgery is because previous research notes that the periods during and immediately following surgery are particularly distressing for parents of children with CHD (Franck et al., 2010).
- Participants must be able to comprehend and speak English, because there are limited funds and it is, therefore, not possible to accommodate translation or interpretation services.

Participants will be excluded if:
- Their child is deceased
- Their child is hospitalised at the time of interview
- Their child has undergone heart transplant

The rationale for these exclusion criteria is that each represent a separate, unique experience, which have different challenges to those being investigated in the proposed research.

**4. How will participants be recruited and from where? Be as specific as possible. Ensure that you provide the *full versions* of all recruitment materials you intend to use with this application (e.g., adverts, flyers, posters).**
Participants will be recruited online via heart charities. Relevant charities have been contacted to establish interest and ability to support with recruitment. To date, one national charity has agreed to involvement and to advertise the study once it has gained ethical approval. Charities agreeing to involvement will be sent several materials to advertise the study. Firstly, for social media and website advertisements the charities will be sent a link to the participant information sheet (Appendix A) and an image of the study advertisement (Appendix B). Secondly, charities will be sent a covering email (Appendix C) to ask if they are able to advertise the study via their email lists. For this purpose, charities will be sent a set text to include in the email (Appendix D) and a PDF version of the participant information sheet to attach.

The lead researcher will create separate Twitter and Facebook pages for the sole purpose of advertising the study. Due to uncontrollable site settings, the Facebook page may link to the researcher's personal profile, which uses the strictest closed privacy settings and will not directly be used for study advertisement. The same social media advertisement and link to participant information sheet will be used. The researcher will 'tweet'/'post' about the study, inviting eligible individuals to take part. Active charities and groups for parents for CHD in the UK will be contacted and asked to 'retweet''/'repost' the advertisement on their page or group. Asking groups to post on behalf of the lead researcher will support anonymity by encouraging individuals to email the researcher about the study, rather than posting responses on an open forum. Furthermore, posts made by the lead researcher will state that the study can only be discussed via email.

The participant information sheet will contain the university email address of the lead researcher, which participants can contact to express interest in taking part or ask any questions. It will be made clear in the information sheet that the email address is not monitored at all times. The lead researcher will respond to expressions of interest by asking individuals to electronically complete and return an expression of interest form (Appendix E), which will ask details related to the inclusion / exclusion criteria. This will allow the individual to be notified quickly if it becomes apparent that they are not eligible to take part in the study. If this is the case, individuals will be informed sensitively via email (Appendix F). The expression of interest form will also ask individuals to detail their current address, to ensure that relevant procedures can be followed in the event of any risk or safeguarding concerns, for example sharing the participant's address with relevant authorities should there be an immediate risk.

Following completion of the expression of interest form, the lead researcher will respond to arrange an interview. Interviews will take place either via Microsoft Teams (which will be recommended) or, if the participant prefers, via telephone or other preferred videoconferencing software (e.g., Skype). An account on alternative videoconferencing software will be set up for the purposes of the research only. It will be made clear in the information sheet, consent form, any email correspondence, and prior to the start of the interview that the security of telephone or videoconferencing software (other than Microsoft Teams) cannot be guaranteed to ensure that participant provide full informed consent to taking part via this medium.

Once an interview is arranged, the lead researcher will send a further copy of the participant information sheet and consent form via email. If applicable, an invite to a Microsoft Teams meeting at the agreed interview date and time and guidance on how to use Microsoft Teams (Appendix G) will also be shared.

Ideally, participants will be recruited to be interviewed individually to allow sufficient time to recall their experiences without interruption or outside influence. However, if there are difficulties with recruitment then couples would be allowed to be interviewed together. Individuals who contact the lead researcher requesting to be interviewed as a couple will receive a response (Appendix H) outlining that the study initially intends to focus on interviewing parents individually, but that this may change. Individuals will be asked if they are happy to be contacted at a later time if this is the case. Alternatively, if the study is over-subscribed then a balance of participants (e.g., equal number of mothers and fathers, or mixture of parents with other children without CHD and parents with an only child) would be sought.

Recruitment is anticipated to close by September 2021, unless the minimum number of participants (n=6) have not been recruited. Any additional individuals who contact the lead researcher to express interest after this point, or after the study has reached full subscription, will be replied to with an email thanking them for their interest and informing them that recruitment to the study has now been closed.

**5. Briefly describe your data collection and analysis methods, and the rationale for their use.**
The study will use qualitative methodology. Data will be collected using semi-structured interviews about parents' experiences of having a child with SVCHD, and any associated impacts upon their identity and / or parental role. The interviews will be conducted via Microsoft Teams (which will be recommended) or, if participants prefer, via telephone or other videoconferencing software. Interviews will be audio-recorded. Interview questions will be guided by the research aims, existing research literature, and feedback from the research supervisors and a national CHD charity. Semi-structured interviews allow flexibility in collecting rich and meaningful data and are recommended

when using Interpretative Phenomenological Analysis (IPA), which will be the method of analysis in this research.

The study will use IPA to analyse data (Smith et al., 2009). Unlike other qualitative approaches (e.g., thematic analysis or grounded theory), IPA is an idiographic, phenomenological method seeking to understand experiences and meaning making, and associated psychological entailments, for individuals and groups. It pays particular attention to patterns and divergences within and across accounts for small samples. This will allow fulfilment of the aim to explore individual meaning of role and identity when parenting a child with SVCHD, which could not be achieved by other qualitative approaches. For example, thematic analysis would only allow exploration of themes across the whole data set. Furthermore, IPA has proven insightful with regards to the topic of identity (Smith, 2004) and health research (Smith, 2011). All of these features make IPA an ideal method for the current research.

**6. What plan is in place for the storage, back-up, security and documentation of data (electronic, digital, paper, etc.)? Note who will be responsible for deleting the data at the end of the storage period. Please ensure that your plans comply with General Data Protection Regulation (GDPR) and the (UK) Data Protection Act 2018.**
For the duration of the research, all data will be stored electronically on the university's secure, encrypted sever (Lancaster University VPN) or in the university's approved, secure cloud storage. Electronic participant identifiable information (e.g., expression of interest forms) will be kept in a password-protected file, separate from anonymised transcripts. This data will be deleted following examination of the thesis.

Upon completion of the research, all other data, including typed interview transcripts, will be transferred to the Doctorate in Clinical Psychology research administration team using a secure university-approved procedure. The data will be retained for a period of ten years. Following this retention period, the data will be deleted by the administration team under the supervision of the research supervisor.

**7. Will audio or video recording take place?** ☐ no ☒ audio ☐ video
**a. Please confirm that portable devices (laptop, USB drive etc) will be encrypted where they are used for identifiable data. If it is not possible to encrypt your portable devices, please comment on the steps you will take to protect the data.**
Regardless of the interview format (e.g., Microsoft Teams, telephone, Skype), all interviews will be audio recorded using a digital audio recorder provided by the Lancaster Doctorate in Clinical Psychology programme. No video recording will take place. The digital audio recorder cannot be encrypted. As such, data will then be transferred, as soon as practicable following completion of each interview, to university's secure, encrypted sever (Lancaster University VPN) or in the university's approved, secure cloud storage. The recording will then be immediately deleted from the digital recorder. It is expected that this process should occur immediately following completion of the interview. However, in the event that there is a delay in this process, the recorder will be kept with the researcher at all times until upload of the data is successful. Interview recordings will be deleted from the secure storage once transcription is complete.

**b What arrangements have been made for audio/video data storage? At what point in the research will tapes/digital recordings/files be destroyed?**
Audio recordings of interviews will be stored on the university's secure, encrypted sever (Lancaster University VPN) or in the university's approved, secure cloud storage until transcription has been completed. Once transcription has been completed and checked, the recordings of interviews will be deleted. Audio recordings of verbal consent provided by participants will be retained by the

Doctorate in Clinical Psychology research administration team for a period of ten years. These will be stored separately from other data associated with the research, including transcriptions of interviews.

**Please answer the following questions *only* if you have not completed a Data Management Plan for an external funder**

**8a. How will you share and preserve the data underpinning your publications for at least 10 years e.g. PURE?**

Data will be transferred electronically to the Doctorate in Clinical Psychology research administration team using a secure, university-approved procedure. It will then be stored electronically on the university's secure, encrypted sever for a period of 10 years, as per usual Doctorate in Clinical Psychology course procedures.

**8b. Are there any restrictions on sharing your data?**

Due to the small sample size within the research, there is a risk that participants may be identified from their interviews, even after full anonymisation. As such, supporting data will only be shared on request via data repository and access will be granted on a case-by-case basis by the Faculty of Health and Medicine, via the research supervisor.

**9. Consent**

**a. Will you take all necessary steps to obtain the voluntary and informed consent of the prospective participant(s) or, in the case of individual(s) not capable of giving informed consent, the permission of a legally authorised representative in accordance with applicable law?** yes

**b. Detail the procedure you will use for obtaining consent?**

Informed consent will be sought from all participants prior to them taking part in the research. An electronic link to the participant information sheet will be included in all advertisements of the study, so that individuals are able to access and read this before contacting the lead researcher to express interest. Participants will also be able to ask any questions they have about the study via email. All participants will receive further copies of the participant information sheet and consent form (Appendix I) via email for their information once an interview date has been arranged. The process will ensure that participants are fully informed of the purpose of the research and methods used, as well as issues pertaining to confidentiality and their right to withdraw, prior to taking part.

Due to the Covid-19 pandemic, interviews will be conducted via video-conferencing software. Microsoft Teams will be recommended but if participants express preference to take part via telephone or other videoconferencing software then this will be considered. In the event that participants do not answer the telephone or video call at the time of the interview, the researcher will call back twice more, after 5 and 10 minutes. If all calls went unanswered, the researcher would assume that the individual no longer consented to take part and would not persist in trying to contact them.

At the beginning of the scheduled interview, participants will have a further opportunity to ask any questions they may have about the study. Consent to participate will be established verbally, immediately prior to the interview, as each item on the consent form will be read out and verbally agreed to in turn before the interview is able to commence. This verbal consent will be recorded separately to the recording of the interview.

**10. What discomfort (including psychological e.g., distressing or sensitive topics), inconvenience or danger could be caused by participation in the project? Please indicate plans to address these**

**potential risks. State the timescales within which participants may withdraw from the study, noting your reasons.**

The research involves asking participants to talk about personal experiences in relation to having a child with SVCHD, a serious and life-threatening heart condition, which could be a sensitive and / or upsetting topic for some. Participants will be fully aware of the intended topics of discussion during the interview, as they will have access to the information sheet prior to taking part. Information regarding appropriate sources of support will be included in the information sheet. At the end of the interview a debriefing conversation will take place, during which participants will be asked how they found the interview and how they feel. Participants will also be provided with a debriefing sheet via email following the interview (appendix J).

In the event that a participant becomes distressed, the interview will be stopped, and the participant allowed as much time as needed to recover and to make an informed decision about whether they would like to continue with the interview. They would be under no pressure to do so. During this time, recording of the interview would also be stopped. As interviews will take place remotely, I will not be in the room with the participants. I will therefore use my clinical skills to assess, respond to and contain distress, for example active listening. Having recently completed a remote working placement within an adult mental health service, I would draw upon my experience of remotely supporting individuals experiencing distress. I would also ask the participant what support they have in place, for example support from family members or friends.

In the event that risk or safeguarding concerns arise regarding risk to self, I would explore this risk, including thoughts, plans and intent. If I felt concerned about a participant's safety, I would work with them to develop a safety plan, including speaking to a trusted person, their GP or, if necessary, calling emergency services or attending their local A&E department. If immediate concerns were to arise, participants would have provided their address on the expression of interest form, and this would be shared with relevant authorities.

In the event that risk or safeguarding concerns arise in relation to the wellbeing of a child or vulnerable adult, or in relation to the professional behaviour from a member of staff working within services, the Lancashire and South Cumbria NHS Foundation Trust (LSCFT) relevant safeguarding policies (LSCFT, 2019; 2020) would be followed. These policies state to seek advice from the LSCFT Safeguarding Advice and Consultancy Service (01772 777153) in the first instance. I would discuss my intention to do so, and reasons for this, with the participant wherever possible and safe to do so. In accordance with LSCFT (2019; 2020) safeguarding policies, if immediate concerns were to arise, as above, I would share information with relevant authorities including, where appropriate, calling 101 or 999.

With regards to any risk or safeguarding concerns, I would also contact both of my research supervisors, one of whom is a qualified Clinical Psychologist, to ensure that all risk management processes have been followed correctly and that there is no further support that I could have provided. In the event that both research supervisors are unavailable, I would contact one of the three Doctorate in Clinical Psychology course directors, who are all qualified Clinical Psychologists. In the first instance, this would be the Research Director, Dr Ian Smith.

Participants will have the option to withdraw from the study at any time before or during the interview without giving any reason. Once the interview has been completed, participants are welcome to withdraw their data up to two weeks after the interview. The reason for this time limit is that the data will then begin to be transcribed, analysed and incorporated into themes.

**11. What potential risks may exist for the researcher(s)? Please indicate plans to address such risks (for example, noting the support available to you; counselling considerations arising from the sensitive or distressing nature of the research/topic; details of the lone worker plan you will follow, and the steps you will take).**
Risks to the research will be minimised as interviews will take place remotely. The email address available for participants to contact to express interest in the research will be a university email address. The same university email address will also be used to set up meetings on Microsoft Teams or other videoconferencing software. Any telephone calls made will use the 'unknown number' feature, which would not show the lead researcher's caller ID. I have agreed regular supervision with my research and field supervisor, as part of a thesis contract, and I would use this space to explore any issues, for example if the content of an interview had been particularly emotive.

**12. Whilst we do not generally expect direct benefits to participants as a result of this research, please state here any that result from completion of the study.**
There will not be any direct benefits to participants as a result of this research. However, they may find having the opportunity to share their experiences and stories positive. Participants will also have the opportunity to request to receive a summary of the research findings, should they wish to. In addition, is it hoped that the results of the research might help healthcare professionals working with parents of children with SVCHD to better understand parents' experiences and, in turn, improve the support offered.

**13. Details of any incentives/payments (including out-of-pocket expenses) made to participants:**
The study will not offer any financial incentives. Due to the current Covid-19 pandemic, interviews will be conducted via free virtual teleconferencing software or telephone. As such, no expenses should be incurred. To ensure this, any participants choosing to be interviewed via telephone will be telephoned by the researcher.

**14. Confidentiality and Anonymity**
**a. Will you take the necessary steps to assure the anonymity of subjects, including in subsequent publications?** yes
**b. Please include details of how the confidentiality and anonymity of participants will be ensured, and the limits to confidentiality.**
All reasonable steps will be taken to protect the anonymity of participants involved in the study, for example transcriptions of audio recordings will use pseudonyms. The interview will not require participants to name their child's hospital of treatment, however if participants do mention this it would be excluded from interview transcripts. The research supervisors will not review transcripts until they have been anonymised. Themes generated through analysis will represent the entire sample rather than specific, identifiable participants. Anonymous, direct quotes may be used in reports or publications from the study, but these quotes will not be attributed to the name of participants. Care will also be taken not to include any direct quotations that may contain easily identifiable information.

The limits of confidentiality will be outlined on the information sheet and consent form. The lead researcher would only break confidentiality if there were concerns regarding risk of harm coming to the participant or someone else, or if there were safeguarding concerns regarding a child, vulnerable adult or the behaviour of a professional. If this case arises, then the concept of confidentiality will be revisited with the participant, and the interview will be terminated. The concern would be reported to the research supervisors and shared with relevant agencies.

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

As part of the development of the research protocol, a national CHD charity viewed and provided feedback on the participant materials (information sheet, consent form, advertisement, debrief sheet) and the interview schedule). The charity provided feedback on the language used and the appropriateness of questions asked.

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

- Anonymised transcripts will be seen by the research supervisors (Dr Craig Murray and Dr Anja Wittkowski) to check and support with coding and the generation of themes.
- The study will be submitted as part of the doctoral thesis required for the Doctorate in Clinical Psychology.
- All participants will be asked if they would like to receive a summary once it has been written up and examined. Any charity involved in supporting the research and any individuals who expressed interest in the study, but were not able to take part, will also have the same opportunity.
- If deemed suitable, and with support of both the research and field supervisor, results of the research may be submitted for publication in a relevant, peer-reviewed journal.
- The research will be presented to all year groups, research and clinical staff of the DClinPsy programme in Summer 2022.
- It is possible that, should the research be deemed of interest, the findings may be presented at a future conference.

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

- The researcher (SD) has previous experience working as an Assistant Psychologist in a Cardiology team at a tertiary children's hospital and, as such, will need to remain aware of their own perspectives throughout the duration of the research. This will be managed through the use of supervision and by keeping a reflective journal, as is standard when using IPA.
- The use of videoconferencing software to conduct interviews could present challenges to developing rapport with participants, and it is important to recognise the limitations of this. The lead researcher has recent experience of completing an Adult Mental Health clinical placement on the Doctorate in Clinical Psychology remotely, using only telephone or videoconferencing software, so will seek to use the skills developed from this experience when conducting interviews with participants. This will also be managed through regular supervision and a reflective journal to note any thoughts or feelings associated with the interview process or how this may have differed from a face-to-face interview.
- The ongoing Covid-19 pandemic is likely to have a significant impact of children with CHD and their families. Children with CHD are particularly vulnerable to infection, so are considered to fall in the 'high risk' category for Covid-19. Those with SVCHD are at even higher risk due to the complexity of their condition. As such, these children and their families are likely to have spent long periods of time 'shielding' since the Covid-19 outbreak in the UK and may have experienced various psychological impacts. For example, parents may feel particularly anxious about their child's wellbeing and health. Furthermore, given the uncertainty regarding the future prevalence and progression of Covid-19, it is possible that further restrictions or lockdown measures may be applied in the UK in future. This may impact psychological wellbeing of children with CHD and their families and, in turn, may affect recruitment to the study.

**SECTION FOUR: signature**

**Applicant electronic signature**: Shannon Dandy Date 16/10/2020

Student applicants: please tick to confirm that your supervisor has reviewed your application, and that they are happy for the application to proceed to ethical review ☒

**Project Supervisor name** (if applicable): Dr Craig Murray Date application discussed 13/10/2020

**Submission Guidance**

1. **Submit your FHMREC application by email to Becky Case (**fhmresearchsupport@lancaster.ac.uk) as two separate documents:
    i. **FHMREC application form.**
    Before submitting, ensure all guidance comments are hidden by going into 'Review' in the menu above then choosing *show markup>balloons>show all revisions in line.*
    ii. **Supporting materials.**
    Collate the **following materials for your study, if relevant, into a single word document:**
        a. **Your full research proposal (background, literature review, methodology/methods, ethical considerations).**
        b. Advertising materials (posters, e-mails)
        c. Letters/emails of invitation to participate
        d. Participant information sheets
        e. Consent forms
        f. Questionnaires, surveys, demographic sheets
        g. Interview schedules, interview question guides, focus group scripts
        h. Debriefing sheets, resource lists

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

2. Submission deadlines:
    i. Projects including direct involvement of human subjects **[section 3 of the form was completed]**. The *electronic* version of your application should be submitted to Becky Case **by the committee deadline date.** Committee meeting dates and application submission dates are listed on the FHMREC website. Prior to the FHMREC meeting you may be contacted by the lead reviewer for further clarification of your application. Please ensure you are available to attend the committee meeting (either in person or via telephone) on the day that your application is considered, if required to do so.
    ii. The following projects will normally be dealt with via chair's action, and may be submitted at any time. **[Section 3 of the form has *not* been completed, and is not required].** Those involving:
        a. existing documents/data only;
        b. the evaluation of an existing project with no direct contact with human participants;
        c. service evaluations.

3. **<u>You must submit this application from your Lancaster University email address, and copy your supervisor in to the email in which you submit this application</u>**

# Having a child with single ventricle congenital heart disease: Parental experience of role and identity

Research Protocol
Version 4.0

Date: 19th July 2021

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## Introduction

Congenital Heart Disease (CHD) is a structural heart defect which develops in the womb (British Heart Foundation, n.d) and has a global prevalence of approximately 9 in 1000 live births. The prevalence of CHD has increased over time, and it has become one of the most pervasive congenital abnormalities (Bernier et al, 2010; van Der Linde et al, 2011). The severity of CHD ranges from mild, such as a small hole in the heart, to severe, such as parts of the heart being unformed. Most children with CHD must undergo at least one surgical procedure (Hoffman & Kaplan, 2002). Although overall survival rates for children with CHD have improved, with around 90% reaching adulthood (Opić et al, 2016), exact treatment and prognosis varies according to CHD type (Šamánek, 2000). For example, single ventricle CHD (SVCHD) is a severe condition in which part of the heart, usually a lower chamber or inlet valve, does not develop normally (British Heart Foundation, 2010). To improve their circulation, children with this condition must undergo a series of operations, the final being the Fontan surgery. However, all treatment for SVCHD is palliative and mortality rates are high (Kaulitz & Hofbeck, 2005).

Due to the complex nature of living with CHD, children with this condition and their families are at increased vulnerability to psychosocial distress (Franck et al, 2010; Jackson et al, 2015; Soulvie et al, 2012), because they are exposed to various challenging circumstances, such as hospitalisation and surgery. In recognition of the psychological impact of CHD and associated experiences, national guidelines state that psychological support must be available at any stage of care to children and their families or carers (National Institute for Health Care and Excellence, 2017; NHS England, 2016). As such, multi-disciplinary teams (MDT) at large paediatric centres often include clinical psychologists who support children and families

using psychological interventions, in addition to promoting wider psychological understanding through liaison, teaching and supervision with staff (Mercer et al, 2015).

**Psychosocial Impact on Parents**

Parents of children with CHD have been found to be at particular risk of psychological distress in comparison to parents of healthy children. For example, parents of children with CHD have been found more likely to experience anxiety and depression (Alkan et al, 2017; Lawoko & Soares, 2002), parenting stress (Uzark & Jones, 2003) and post-traumatic stress (Kolaitis et al, 2017). Although some studies have found no difference in levels of psychological distress between parents of children with CHD and normative samples (Fischer et al, 2012; Vrijmoet-Wiersma et al, 2009), some have suggested that this may be due to parents experiencing denial about their child's CHD (Nakazuru et al, 2017; Utens et al, 2002). However, similar levels of psychological wellbeing among parents of children with CHD and parents of 'healthy' children may simply suggest that some parents are able to psychologically come to terms with, and adapt to, their child's diagnosis (Jackson et al, 2018; Lumsden et al, 2020; Lumsden et al, 2019).

It is clear that having a child with CHD is a significant life event, regardless of the level of distress experienced by parents as a result. In a qualitative study by Re et al (2013) twenty-six mothers described a range of feelings including shock, concern, guilt, fear and loss of a 'normal' child when receiving a prenatal diagnosis of CHD. Parents also noted a strong sense of responsibility and protectiveness over their unborn child. These feelings have been consistently reported across cultures (Im et al, 2018; Nakazuru et al, 2017).

In addition to managing their own emotional responses, parents of children with CHD face additional challenges that may go some way to explaining their vulnerability to

psychological distress. They are expected to provide support to their child and other family members (Jackson et al, 2015), make crucial decisions about their child's care (Rempel et al, 2004), manage financial instability due to taking sick leave or leaving employment to care for their child (Connor et al, 2010) and adapt to lifestyle changes, such as reducing social contact to decrease potential exposure to illness (Carey et al, 2002). Indeed, stages of care in which these challenges are likely to be more acute have been described as the most significant periods of stress for parents: diagnosis (Bratt et al, 2019; Thomi et al, 2019), hospitalisation (Lisanti et al, 2017; Simeone et al, 2018; Sjostrom-Strand & Terp, 2019) and surgery (Harvey et al, 2013; Re et al, 2013; Wei et al, 2016). For parents of children with more severe CHD, such as SVCHD, challenges such as hospitalisation and surgery are likely to occur more frequently. As such, it has been proposed that parental distress increases according to CHD severity (Solberg et al, 2012).

Although much research has considered levels of psychological distress among parents of children with CHD, a review of 94 studies by Wei et al (2015) identified a lack of qualitative research. Several studies have since sought to address this gap, by qualitatively considering parents' experiences in a number of areas including diagnosis (Thomi et al, 2019), surgery (Wei et al, 2016) and early years (Barreto et al, 2016). More recently, Lumsden et al (2020) sought to understand the parental journey through to adolescence and found that parents were able to establish a sense of normality once they adjusted to and accepted changes that CHD brings to their parental role.

**Parental Role and Identity**

The definitions of role and identity have been subject to much contention. Broadly, identity can be considered to be a set of meanings, such as personal or social, that shape an

individual's understanding of who they are (Burke, 2009), whereas roles have been conceptualised as social positions denoted by how an individual behaves, or is expected to behave (Turner, 2001). The roles that an individual adopts can influence their identity, and vice versa. Becoming a parent, whether with the birth of a first or subsequent child, is a significant life event which leads to changes in identity and development of specific parental roles. A review of 115 articles by Shudy et al (2006) and a further review of 33 articles by Abela et al (2020) found parental role, identity and psychological wellbeing are acutely challenged when the child is critically unwell. This finding has been reported among parents of children with chronic illness, such as chronic kidney disease (Nicholas, 2017) or cancer (Young et al, 2002), as well as among parents whose children have passed away (Toller, 2008).

Indeed, parental role and identity has been touched upon in qualitative studies exploring parents' experiences of having a child with CHD. At diagnosis, parents describe becoming the parent of a 'sick' child, in addition to a heightened sense of parental role, particularly to make decisions (Rempel et al, 2013). Conversely, Simeone et al (2018) found that when their child is admitted to hospital, parents feel stripped of their parental role due to the unfamiliar medical environment, lack of physical contact with their child and personal cares being mostly completed by medical staff. Similarly, fathers have reported feeling loss of purpose and role throughout the journey of parenting a child with CHD (Gower et al, 2017). These experiences of loss of role were associated with distress, helplessness and guilt. In contrast, transitioning home from hospital following surgery has been found to provoke role conflict for parents, as they describe struggling to balance being both parent and nurse to their child (Gaskin, 2018; Harvey et al, 2013). Quantitative research has supported qualitative findings that alterations to parental role and self-identity as a result of having a child with

CHD are associated with parents' experience of distress (Diffin et al, 2016). Whereas, in Lumsden et al's (2020) study, parents reported that they were able to establish normality and a sense of control when they began to accept alterations to their parental role, such as becoming an expert in their child's condition. Parental perception of their own role and identity, therefore, seems to be a significant factor in influencing the psychological wellbeing of parents of children with CHD.

Although the above research contributes to our understanding of parents' experiences of having a child with CHD, each study only touched upon experiences of alterations to parental role and/or identity briefly or as part of a broader picture. No study has yet explored the implications of having a child with CHD on parents' perceptions of their own role or identity in detail. The current study, therefore, aims to explore how parents make sense of their identity and parental role when parenting a child with SVCHD, as well as any associated psychological implications. It is anticipated that the findings will inform our understanding of parents' psychological needs and shape how they are supported by professionals. The study will focus on parents of children with SVCHD as this is a unique population in which parents are more frequently exposed to challenges such as poor prognosis, hospitalisation and surgery. A focus on parents of children with this condition also promotes homogeneity among the sample, as required for Interpretative Phenomenological Analysis (IPA) as the method of analysis. The research questions are as follows:

- How do parents experience their role as a parent when their child has single ventricle CHD?
- How do parents experience their identity when their child has single ventricle CHD?

- What are the psychological implications for parents when they experience changes to parental role and/or identity as a result of their child having single ventricle CHD?

## Methods

### Design

The research will employ a qualitative design, using semi-structured interviews and Interpretative Phenomenological Analysis (IPA) (Smith et al, 2009). Semi-structured interviews provide participants with the opportunity to relay detailed accounts of their experiences whilst allowing the researcher to maintain structure and focus on the research questions, for example, through the use of prompts. When utilising IPA, use of semi-structured interview is the recommended and most common method of data collection (Smith & Osborn, 2008; Smith et al, 2009). IPA is an idiographic, phenomenological method that seeks to understand individuals' experiences, and associated psychological entailments, and the meaning they give to these (Smith et al, 2009). In relation to the proposed study, IPA has proven insightful with regards to the topic of identity (Smith, 2004) and health research (Smith, 2011).

### Participants

The study will recruit 6-12 participants, as a small, defined sample is recommended when using IPA because of its idiographic focus (Murray & Wilde, 2020). The sample will be a convenience sample, but individuals' participation in the study will be subject to meeting both the inclusion and exclusion criteria. The sample will be based in the United Kingdom. Participants will be parents of children with SVCHD who have had their completed Fontan

surgery, which is the last of three stages of surgery in usual treatment for children with this condition. In the first instance, the study will recruit participants as individuals to allow sufficient time to recall their experiences without interruption or outside influence. However, if there are difficulties with recruitment then couples would be allowed to interview together.

***Inclusion Criteria***

Individuals will be eligible to take part in the study if:

- They have a child with SVCHD
- The child had their completed Fontan surgery at least six months prior to the interview
- The child is aged 25 years or under
- They reside in the UK
- They are able to comprehend and speak English

***Notes on Inclusion Criteria***

The rationale for the child needing to have had completed Fontan surgery is that this will allow exploration of the complete surgical journey and promote homogeneity among participants, which is sought when using IPA (Smith et al, 2009). The requirement for the child to be at least six months post-surgery has been set based on previous findings that the time periods during and immediately following surgery are particularly distressing for parents of children with CHD (Franck et al, 2010).

Participants must be able to comprehend and speak English because there are limited funds to conduct the study and it is therefore not possible to accommodate translation or interpretation services.

***Exclusion criteria***

Individuals will be excluded from taking part in the study if:

- Their child is deceased
- Their child is hospitalised at the time of interview
- Their child has undergone heart transplant

The rationale for the above exclusion criteria is that each represent a separate, unique experience and the presence of any of these factors may mean that these parents are facing different challenges to those being investigated by the proposed research.

**Materials**

Prior to the interview, materials to be used in the study include participant information sheet (Appendix A), participant consent form (Appendix I), email advertisement text (Appendix D), social media advertisement (Appendix B), participant expression of interest form (Appendix E) and guidance on using Microsoft Teams (Appendix G).

A semi-structured interview will be used (see Appendix K for an example semi-structured interview topic guide). The interviews will be recorded using a digital recorder.

At the end of the interview, participants will receive a debrief sheet (Appendix J) containing relevant details of further sources of support, should they wish to access it.

**Procedure**

***Recruitment***

Participants will be recruited to participate in the study online via CHD charities based in the UK. Relevant charities have been contacted by the lead researcher, Shannon Dandy, to establish any interest and ability to support with recruitment to the study. To date,

one national charity has agreed to involvement and to advertise the study once it has gained ethical approval. Charities agreeing to involvement will be sent several materials to advertise the study. Firstly, for social media and website advertisements the charities will be sent a link to the participant information sheet and an image of the study advertisement. Secondly, charities will be sent a covering email (Appendix C) to ask if they are able to advertise the study via their email lists. For this purpose, charities will be sent an email advertisement text and a PDF version of the participant information sheet to attach.

The lead researcher will also create separate Twitter and Facebook pages for the sole purpose of advertising the study. Due to uncontrollable site settings, the Facebook page may link to the researcher's personal profile, which uses the strictest closed privacy settings and will not directly be used for study advertisement. The same social media advertisement and link to participant information sheet will be used. The researcher will 'tweet'/'post' about the study, inviting eligible individuals to take part. Active charities and groups for parents for CHD in the UK will be contacted and asked to 'retweet''/'repost' the advertisement on their page or group. Asking groups to post on behalf of the lead researcher will support anonymity by encouraging individuals to email the researcher about the study, rather than posting responses on an open forum. Furthermore, posts made by the lead researcher will state that the study can only be discussed via email.

The participant information sheet will contain the email address of the lead researcher (Shannon Dandy) that participants are able to contact to express interest in taking part or to ask any questions. It will be made clear in the participant information sheet that the email is not monitored at all times. The lead researcher will respond to individuals wishing to participate by asking them to complete and return an expression of interest form, which will ask for details related to the inclusion / exclusion criteria such as the age of their child and

confirmation that their child had completed Fontan surgery at least six months prior. This will allow the individual to be notified quickly if it becomes apparent that they are not eligible to take part in the study. If this is the case, individuals will be informed sensitively via email (Appendix F). The expression of interest form will also ask individuals to detail their current address. This process will ensure that relevant procedures can be followed in the event of any risk of safeguarding concerns; for example, sharing the participant's address with relevant authorities should there be an immediate risk. Following completion of the expression of interest form the lead researcher will contact the individual to arrange an interview.

The study will initially seek to interview participants individually, so individuals who contact the lead researcher requesting to be interviewed as a couple will be informed of this via email (Appendix H). However, as this may change depending on the number of participants recruited, the researcher will ask the individual if they are happy to be contacted at a later date, should interviews of couples become a feature of the study. Alternatively, if the study is over-subscribed, then expression of interest forms would be screened to seek a balance of participants, for example an equal number of mothers and fathers or a mixture of parents with other children who do not have CHD and parents who have an only child with CHD.

Recruitment is expected to close by September 2021, unless the minimum number of participants (n=6) have not been recruited. Any additional individuals who contact the lead researcher to express interest in participating after this point, or after the study has reached full subscription, will be replied to with an email thanking them for their interest and informing that recruitment to the study has now been closed.

***Interviews***

Once an interview date has been arranged, participants will be able to choose how the interview will take place. Microsoft Teams will be the recommended and default format, but if participants express preference to take part via telephone or other videoconferencing software then this will be considered. However, it will be made clear to participants in the information sheet, consent form, any email correspondence and verbally prior to the start of the interview that the security of telephone of videoconferencing software (other than Microsoft Teams) cannot be guaranteed. This will ensure that participants provide full informed consent to taking part via this medium.

Prior to the interview, all participants will receive a further copy of the participant information sheet and consent form for their information. If applicable, an invite to a Microsoft Teams meeting at the agreed interview time and guidance on using Microsoft Teams will also be sent via email. At the beginning of meetings with participants, they will have a further opportunity to ask any questions they may have about the study. Participants will then provide verbal informed consent immediately prior to the interview, with each item on the consent form read out by the lead researcher and agreed to verbally by the participant before the interview commences. This verbal consent will be recorded separately to the recording of the interview.

The meeting will consist of introductions, confirming demographic information and consent, a semi-structured interview, debrief and closing questions from the participants. Prior to commencement of the interview, the lead researcher will recap the information in the participant information sheet and the consent form. As above, each item on the consent form will be read aloud by the researcher and the participant will need to verbally agree to each in turn before the interview can begin. The researcher will audio record the process of gaining

verbal consent. Participants will then be asked to choose a pseudonym that will be used in transcriptions of interviews and ascribed to any direct quotations documented in the report.

The interview will be audio recorded and last for approximately 1-1 ½ hours. The interview will be semi-structured to allow the lead researcher to initially outline the focus of the interview and provide occasional prompts where necessary, but otherwise allow the participant to talk freely. Interview questions and any prompts used will be reviewed after each interview, to allow the lead researcher to respond and adapt future interviews. After the interview, a debriefing conversation will take place when participants will be asked how they found the interview and how they are feeling. Participants will also be asked if they have any questions and will be reminded that they are able to withdraw their data from the study up to two weeks after the interview. Additionally, participants will be asked if they would like to receive a summary of the study after it has been submitted and assessed and, if they say yes, this will be sent to them via email. Finally, participants will be sent, via email, a debriefing sheet immediately after the interview. This will consider any sensitive or distressing topics discussed and will include contact details for relevant support organisations.

**Data Management Plan**

***During the study***

For the duration of the research, all data will be stored electronically on the university's secure, encrypted server (Lancaster University VPN) or in the university's approved, secure cloud storage. Regardless of the interview format (e.g., Microsoft Teams, telephone, Skype), all interviews will be audio recorded using a digital audio recorder provided by the Lancaster Doctorate in Clinical Psychology programme. No video recording will take place. The digital audio recorder cannot be encrypted. As such, recordings will be

transferred to the secure university server or secure university cloud storage as soon as practicable following the interview. The recording will then be deleted from the digital recorder immediately. It is expected that this process should occur immediately following completion of the interview. However, in the event that there is a delay in this process, the recorder will be kept with the researcher at all times until upload of the data is successful. Participants will be able to withdraw their data up to two weeks after the interview. The reason for this time limit is that the data will then begin to be transcribed, analysed and incorporated into themes.

All reasonable steps will be taken to protect the anonymity of participants involved in the study; for example, transcriptions of audio recordings will use pseudonyms and will be stored on the university's secure encrypted server or in the university's approved, secure cloud storage. Interview recordings will be deleted once the transcription and has been completed. Electronic participant identifiable information, i.e., expression of interest forms, will be kept in a password-protected file, separate from anonymised transcripts. This data will be deleted following examination of the thesis.

***Upon completion of final examination***

Upon completion of the research, electronic participant identifiable information will be deleted. All other data, including typed interview transcripts, will be transferred to the Doctorate in Clinical Psychology research administration team using a secure, university-approved procedure. Recordings of verbal consent provided by participants will also be transferred, separately, to the Doctorate in Clinical Psychology research administration team using a secure, university-approved procedure. These will be stored separately from other data associated with the research. All data will be retained for a period of 10 years, as per

usual Doctorate in Clinical Psychology course procedures. Following this retention period, the data will be deleted by the administration team, under the supervision of the research supervisor.

Due to the small sample size within the study, there is a risk that participants may be identified from their interviews, even after full anonymisation. As such, supporting data will only be shared on request via data repository, and access will be granted on a case-by-case basis by the Faculty of Health and Medicine, via the primary research supervisor (Dr Craig Murray).

**Analysis**

IPA will be the method of analysis used in the research, following the processes outline by Murray & Wilde (2020). Unlike other qualitative approaches (e.g., thematic analysis or grounded theory), IPA is an idiographic, phenomenological method seeking to understand experiences and meaning making, and associated psychological entailments, for individuals and groups. When guided by IPA, the lead researcher pays particular attention to patterns and divergences within and across accounts for small samples. For the proposed study, this will allow fulfilment of the aim to explore individual meaning of role and identity when parenting a child with SVCHD, which could not be achieved by other qualitative approaches. For example, thematic analysis would only allow exploration of themes across the whole data set. It is anticipated, due to time constraints associated with the DClinPsy thesis, that data collection and analysis will be conducted concurrently.

When completing the data analysis, the lead researcher will firstly transcribe interviews verbatim. Notations will then be made and transformed into emergent themes for each transcript individually. Once this process has been completed for all transcripts

separately, analysis will be merged across transcripts to identify themes across the sample. Throughout the process of conducting interviews and analysis, the lead researcher will keep a reflective journal allowing documentation of any feelings, responses or preconceptions to data that may influence the interpretation. This will be discussed with the research supervisors, when necessary.

## Practical Issues

### *Confidentiality and Anonymity*

The digital recorder used to record interviews cannot be encrypted. As such, data will be transferred, as soon as practicable following completion of each interview, to the university's secure, encrypted server or in the university's secure cloud storage. The recording will then be immediately deleted from the digital recorder. All reasonable steps will be taken to protect the anonymity of participants, for example transcriptions of audio recordings will use pseudonyms. Although the interview will not ask participants to state their child's hospital of treatment, if this is mentioned during the interview it will be excluded from transcripts. The research supervisors will not review transcripts until they have been anonymised. Anonymous, direct quotes may be used in reports or publications from the study, but these quotes will not be attributed to the name of participants. Care will also be taken not to include any direct quotations that may contain easily identifiable information.

However, there are limits to confidentiality, which will be outlined on the information sheet and consent form. Specifically, if something said during the interview indicates that the participant or another person may be at risk of harm, or if safeguarding concerns regarding a child, vulnerable adult or the behaviour of a professional arose, then the lead researcher

would need to share this information with relevant agencies. The lead researcher would discuss their intention to do so, and reasons for this, with the participant wherever possible.

### *Expenses*

Participants will be interviewed using free videoconferencing software or telephone. As such, there will be no expenses associated with the study. To ensure this, any participants choosing to be interviewed via telephone will be telephoned by the researcher.

### *Covid-19*

As a result of the ongoing Covid-19 pandemic in the UK, including current restrictions on meeting with others outside of an individual's own household, interviews with participants will be conducted remotely. This may present practical issues; for example, participants may not feel confident using technology or may experience difficulties with internet connection. To minimise such difficulties, Microsoft Teams will be the recommended medium for interviews as the lead researcher is familiar with the software. Where applicable, guidance on how to access and use Microsoft Teams will also be sent to participants in advance of the interview.

## Ethical Considerations

### *Informed Consent*

Informed consent will be sought from participants prior to them taking part in the research. A link to the participant information sheet will be included in all advertisements of the study, so that participants are able to access and read this before contact the lead researcher to express interest in taking part. All participants will receive further copies of the

participant information sheet and consent form for their information via email once an interview date has been arranged. The process will ensure that participants are fully aware of the purpose of the research and methods used, as well as issues pertaining to confidentiality and their right to withdraw, prior to taking part.

Due to the ongoing Covid-19 pandemic, interviews will be conducted remotely. As such, participants will be sent a copy of the consent form via email prior to the interview for their information, but consent will be established verbally. Immediately prior to the interview, each item on the consent form will be read out and verbally agreed to before the interview is able to commence. This verbal consent will be recorded separately to the recording of the interview.

In the event that participants do not answer the telephone or video call at the time of the interview, the researcher will call back twice more, after 5 and 10 minutes. If all calls went unanswered, the researcher would assume that the individual no longer consents to take part and would not persist in trying to contact them.

***Distress and Safeguarding***

The research involves asking participants to talk about personal experiences in relation to having a child with SVCHD, a serious and life-threatening heart condition. This could be a sensitive and/or upsetting topic for some participants. Participants will be fully aware of the intended topics of discussion during the interview, because they will have been informed via the information sheet prior to taking part. Information regarding appropriate sources of support will be included in the information sheet. At the end of the interview, a debriefing conversation will take place when participants are asked how they found the

interview and how they feel. Participants will also be provided with a debriefing sheet via email following the interview.

In the event that the participant becomes distressed, the interview will be stopped, and the participant will be allowed as much time as needed to recover and to make an informed decision about whether they would like to continue. They would be under no pressure to do so. During this time, recording of the interview will also be stopped. As interviews will take place remotely, I as the lead researcher will not be in the room with participants. I will therefore use my clinical skills to assess, respond to and contain distress, for example through active listening. Having recently completed a remote placement within an adult mental health service, I would seek to draw upon my experience of remotely supporting individuals experiencing distress. I would also ask the participant what support they have in place, for example support from family members or friends.

In the event that any risk or safeguarding concerns arise regarding risk to self, I would assess and explore the risk, including thoughts, plans and intent of harm. If I felt concerned about the participant's safety, I would work with them to develop a safety plan. This plan would include the participant speaking to a trusted person, their GP or, if necessary, calling emergency services or attending their local A&E department. If immediate risk were to arise, participants would have provided their address on the expression of interest form, and this would be shared with relevant authorities.

Similarly, should risk or safeguarding concerns arise in relation to the wellbeing of a child or vulnerable adult, or in relation to the professional behaviour from a member of staff working within services, the Lancashire and South Cumbria NHS Foundation Trust (LSCFT) relevant safeguarding policies (LSCFT 2019, 2020) would be followed. These policies state to seek advice from the LSCFT Safeguarding Advice and Consultancy Service (01772

777153) in the first instance. I would discuss my intention to do so, and reasons for this, with the participant wherever it is possible and safe to do so. In accordance with LSCFT (2019; 2020) safeguarding policies, if immediate concerns were to arise I would, as above, share information with the relevant authorities including, where appropriate, calling 101 or 999.

In addition, should any risk or safeguarding concern arise, I would also contact both of my research supervisors, one of whom is a qualified clinical psychologist, to ensure that all risk management processes have been followed correctly and that there is no further support that I could have provided. In the event that both research supervisors are unavailable, I would contact one of the three Doctorate in Clinical Psychology course directors, who are all qualified clinical psychologists. In the first instance, this would be the research director, Dr Ian Smith.

***Participant Withdrawal***

Participants will have the option to withdraw from the study at any time before or during the interview, without giving any reason. In the event that participants do not answer the telephone or video call at the time of the interview, the researcher will call back twice more, after 5 and 10 minutes. If all calls went unanswered, the researcher would assume that the individual no longer consents to take part and would not persist in trying to contact them. Once the interview has been completed, participants will be able to withdraw their data up to two weeks after the interview. The reason for this time limit is that the data will then begin to be transcribed, analysed and incorporated into themes. As such, it will not be possible to identify and withdraw the data.

***Study Oversubscription***

In the event that the study is oversubscribed, the lead researcher will sensitively explain this to interested individuals. The lead researcher would explain that the study is oversubscribed and that a careful process has been used to ensure a homogenous, balanced sample. Individuals would be thanked for their interest and would be offered the opportunity to receive a summary of results once the research has been submitted and assessed.

***Covid-19***

The ongoing Covid-19 pandemic is likely to have a significant impact on children with CHD and their families. Children with CHD are particularly vulnerable to infection, so are considered 'high risk' for Covid-19. Those with SVCHD are at even higher risk due to the complexity of their condition. As such, these children and their families are likely to have spent long periods of time 'shielding' since the Covid-19 outbreak and may have experienced various psychological impacts as a result. For example, parents may feel particularly anxious about their child's wellbeing and health. Furthermore, given the uncertainty regarding the future prevalence and progression of Covid-19, it is possible that further restrictions or lockdown measures may be introduced in the UK. This may impact psychological wellbeing of children with CHD and their families and, in turn, may affect recruitment to the study.

***Stakeholder Involvement***

The participant information sheet, consent form, study advertisement (both social media and email) and interview schedule have been shared with a national CHD charity who have agreed to be involved in the research. The charity reviewed and provided feedback on these materials.

**Dissemination Strategy**

The research will be the thesis of the lead researcher, Shannon Dandy, submitted in partial fulfilment of the Doctorate in Clinical Psychology at Lancaster University. The lead researcher will also present the research to all year groups, research and clinical staff of the DClinPsy programme in Summer 2022. Once the study has been completed and assessed, attempts to publish the research in a relevant academic journal will be made. The research may also be presented at future relevant conferences, should the findings be of interest. Additionally, the findings will be summarised, using lay language, and shared with the charities involved in supporting the research and with any participants who requested a summary.

**Estimated Timescale**

| | |
|---|---|
| November 2020: | Submit for ethical review |
| February - March 2021: | Begin recruitment and interviewing of participants |
| April 2021: | Begin data analysis |
| September 2021: | Recruitment and interviewing ends |
| November 2021: | Finalise data analysis |
| December 2021-March 2022: | Final write up and submission of DClinPsy Thesis |
| August 2022: | Summary of the research to be sent to charities involved and participants if requested |
| September 2022: | Submit for publication |

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**Appendix 4-A**
**Participant information sheet**

# Participant Information Sheet

## *Having a child with single ventricle congenital heart disease: Parental experience of role and identity*

My name is Shannon Dandy and I am a trainee clinical psychologist. I am conducting this research as a student in the Doctorate in Clinical Psychology Programme at Lancaster University, Lancaster, United Kingdom. Thank you for taking the time to read this information sheet. **If, at the end of reading the information, you would like to take part or would like further information, please contact me at s.dandy@lancaster.ac.uk. Please note that this email address is not monitored at all times.**

### What is the study about?

The purpose of this study is to interview parents of children aged 16 years or under with single ventricle congenital heart disease who have had their completed Fontan surgery. Previous research has recognised the unique experiences parents have when their child has single ventricle congenital heart disease and has suggested that there may be an impact on how parents understand their own identity and roles. However, no study has yet considered this in detail. Therefore, the current study is looking to address this gap and provide a platform for parents to speak about their experiences.

### Why have I been approached?

You have been approached because the study is looking to hear about the experiences of people who have a child aged 16 years or under with single ventricle congenital heart disease who had their completed Fontan surgery at least six months ago.

### Are there any requirements to taking part?

Yes. To take part, you should:

- Have a child aged 16 years or under with single ventricle congenital heart disease, who had their completed Fontan surgery at least 6 months ago.
- Live in the UK.
- Be able to understand and speak English. The reason for this is that there is limited funding for the research and, therefore, it is not possible to fund translation or interpretation services.
- Be available to take part in an interview during working hours (Monday – Friday, 9am-5pm).

If your child is currently in hospital or has had a heart transplant it will not be possible to take part in this research. This is because these are unique experiences that will not be able to be fully explored by the current study.

### Do I have to take part?

No. It's completely up to you to decide whether or not you take part and you are under no obligation to do so.

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

If you decide you would like to take part, you would be asked to take part in an interview with me will last for approximately one to one and a half hours. The interview will take place at a time that suits you, between the hours of 9am-5pm from Monday to Friday. Interviews will be audio recorded and carried out remotely. It is recommended that interviews take place via Microsoft Teams and information on how to access and use this software will be supplied prior to the interview, via email. However, if you have a preference for telephone or other videoconferencing software then this would be considered. Please note that the security of interviews completed via telephone or other videoconferencing software cannot be guaranteed due to the nature of the platforms.

During the interview, you will be asked about your experiences of parenting a child with single ventricle congenital heart disease. These experiences will be unique to you, but specific questions will include asking about your journey with your child, your perception of your parental roles, your sense of identity and your psychological wellbeing.

If you do not answer the telephone or video call at the specified time of the interview, the researcher will try to call you twice more, after 5 and 10 minutes. If all calls go unanswered, the researcher will assume that you no longer wish to take part and will not persist in trying to contact you.

## Can I change my mind?

Yes. You can choose to withdraw from the study at any time before or during the interview without giving any reason. Once the interview has been completed, you will be able to withdraw your data for up to two weeks after the interview. You can withdraw from the study by contacting me directly via email. After this time, the data will begin to be anonymised, transcribed, analysed and incorporated into themes. As a result, it will not be possible to identify and withdraw the data.

## Will my data be Identifiable?

Any personal data that you provide will be kept confidentially and separately from your interview responses. The audio recording of your interview will be typed and made anonymous by removing any identifying information, including your name and your child's name. The interview will not ask you to state your child's hospital of treatment, but if this is mentioned during conversation then it will be removed from the typed transcript. Anonymised direct quotations from your interview may be used in the write-up of the study, but a pseudonym (false name) will be used so your name will not be attached to them. All reasonable steps will be taken to protect your anonymity.

The data collected for this study will be stored securely. Only the lead researcher, Shannon Dandy, will have access to participant interviews, which will be stored electronically on a secure drive. The research supervisors, Dr Craig Murray and Dr Anja Wittkowski, will have access to anonymised interview transcripts. Audio recordings of interviews will be deleted once they have been transcribed. At the end of the study, electronic copies of anonymised interview transcripts will be kept securely for 10 years at Lancaster University, in line with university policy. Audio recordings of you providing verbal consent will be kept securely and

separately for the same 10-year period. At the end of this period, the anonymised transcripts and audio recordings of verbal consent will be deleted.
There are some limits to confidentiality; specifically, if something is said in the interview that makes me think that you, or someone else, may be at risk of harm then I will have to break confidentiality and speak to another member of staff or other relevant agencies about this. If possible, I will tell you if I have to do this.

## What will happen to the results?

The results will be summarised and reported in a thesis paper that will be assessed as part of my Doctorate in Clinical Psychology qualification. It may be decided to submit the report to a peer-reviewed journal which, if published, would be accessible via the internet. You will be offered a summary of the research findings have been written, to see how your input contributed to the findings. When, or if, direct quotes from your interview are used in the report or publication, this will be anonymised.

For more information on how Lancaster University processes personal data for research purposes and your data rights, please visit our webpage: www.lancaster.ac.uk/research/data-protection

## Are there any risks?

There are no risks anticipated with participating in this study. However, I am aware that having a child with single ventricle congenital heart disease can be a challenging experience and it may be upsetting to talk about these experiences. Therefore, if you experience any distress during or following participation in the study you are encouraged to let me know. In addition, you will be provided with a debrief sheet that will provide contact details for resources that you could access for support.

## Are there any benefits to taking part?

There are no direct benefits to you for taking part. However, it is hoped that findings from the research might help healthcare professionals working with children with single ventricle congenital heart disease to better understand parents' experiences.

## Who has reviewed the project?

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

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

If you would like to participate in this research or have any further questions, please contact the main researcher on the details below. However, please note that the email is not monitored at all times.

Shannon Dandy
Doctorate in Clinical Psychology
Lancaster University, Lancaster
LA1 4YG
Email: s.dandy@lancaster.ac.uk

You can also contact the main research supervisor if you wish to:
Dr Craig Murray
Doctorate in Clinical Psychology
Lancaster University, Lancaster
LA1 4YG
Email: c.murray@lancaster.ac.uk

**Complaints**
If you wish to make a complaint or raise concerns about any aspect of this study and do not want to speak to the researcher, you can contact:
Dr Ian Smith
Research Director
Doctorate in Clinical Psychology
Lancaster University, Lancaster
LA1 4YG
Email: ian.smith@lancaster.ac.uk

If you wish to speak to someone outside of the Doctorate in Clinical Psychology Programme, you may also contact:
Dr Laura Machin Tel: +44 (0)1524 594973
Chair of FHM REC Email: l.machin@lancaster.ac.uk
Faculty of Health and Medicine (Lancaster Medical School)
Lancaster University
Lancaster
LA1 4YG

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

**Resources in the event of distress**
As part of the interview, you may choose to talk about things that are difficult or upsetting for you and cause distress. These feelings may go away within a few minutes, hours or days. However, should you continue to feel distress, either as a result of taking part or in the future, please contact your GP. The following sources of support may also be useful:

- Your child's Cardiology team (sometimes there is also a Clinical Psychologist who works within the Cardiology team who may be able to offer support).
- Little Hearts Matter: A national charity supporting children with single ventricle congenital heart disease and their families
    - 0121 455 8982
    - Lhm.org.uk
- Children's Heart Federation: A national charity for children with heart conditions and their families
    - 0300 561 0065
    - Chfed.org.uk
- Other sources of emotional support may also be helpful, for example the Samaritan's Helpline: 116 123

**Appendix 4-B**
**Social media advertisement**

Text:
Are you the parent of a child with single ventricle congenital heart disease? Opportunity to be involved in research here: (link to participant information sheet).

Image:

**Appendix 4-C**
**Covering email to charities**

Dear (name)

Thank you very much again for agreeing to be involved in advertising the research study 'Having a child with single ventricle congenital heart disease: Parental experience of role and identity'.

As you may be aware from our previous correspondence, I will be looking to interview parents of children with single ventricle congenital heart disease about their experiences, specifically related to how they perceive their own role and identity. These interviews will take via Microsoft Teams with myself. The interviews will be audio recorded and later transcribed with all identifying information anonymised.

As agreed, I am emailing to confirm that the study has received ethical approval and I am therefore hoping that you could now begin to advertise the study via your website and / or social media. I would be extremely grateful if this is possible, and I have attached the advertisement and an electronic link to the participant information sheet to include in any posts. As per the ethical approval, the posts should state "Are you the parent of a child with single ventricle congenital heart disease? Opportunity to be involved in research here: (*insert link to participant information sheet*)" and have the study advertisement attached as a photo.

Additionally, I wondered if it might be possible to contact anyone on your emailing list who may be interested. For this purpose, I have also attached the approved written text to include in the email and a PDF copy of the participant sheet which can be attached. Your advertisement of the study, in any way, is greatly appreciated.

I look forward to your reply and if you have any further questions, please do not hesitate to contact me. Thank you once again for your support with this research.

Best wishes,

Shannon Dandy
Trainee Clinical Psychologist
Lancaster University

**Appendix 4-D**
**Text to be included in email advertisements**

Dear member,

Are you the parent of a child aged 16 years or under who has single ventricle congenital heart disease and had their completed Fontan surgery at least 6 months ago? If so, we are getting in touch to let you know about a new research opportunity. Information from the lead researcher is detailed below.

Hello,

My name is Shannon Dandy, and I am a trainee clinical psychologist. I am conducting some research as a student in the Doctorate in Clinical Psychology Programme at Lancaster University, Lancaster, United Kingdom.

I am researching the experiences of parents who have a child with single ventricle congenital heart disease, particularly any impact this may have on their understanding of their own role and identity.

The study will involve an interview with me, where you would be asked about your parenting experiences and your understanding of your own role and identity. Interviews will take place remotely via Microsoft Teams. Data from the study will be anonymised, with a pseudonym (fake name) used for any direct quotations in the write-up.

Further information about the study is attached to this email. If you are interested in taking part in the study or have any further questions, then please do not hesitate to contact me at **s.dandy@lancaster.ac.uk.**

I look forward to hearing from you.

Shannon Dandy
Trainee Clinical Psychologist
Lancaster University

**Appendix 4-E**
**Expression of interest form**

**Study Title:** Having a child with single ventricle congenital heart disease: Parental experience of role and identity.

Thank you for expressing an interest in taking part in the above study. In order to ensure that the study is appropriate for you, please fill out the information below and return the form to the lead researcher, Shannon Dandy, via email (s.dandy@lancaster.ac.uk). Shannon will then be in touch to arrange an interview.

**Information about you**

**Name:**

**Age:**

**Gender:**

**Address:**

**Email Address:**

**Telephone Number:**

**Information about your child**

**Child's Age:**

**Child's Gender:**

**When did your child have their completed Fontan surgery?**

**Is your child currently in hospital?**

**Has your child had a heart transplant?**

**I confirm that I am interested in taking part in the above study and would like to be contacted via email by the lead researcher to arrange an interview.**

**Signed:**

**Date:**

**Appendix 4-F**
**Email response to individuals who are not eligible to take part**

Dear (name)

Thank you very much for expressing your interest in the research I am conducting regarding experiences of parents of children with single ventricle congenital heart disease. I really appreciate you taking the time to contact me and to complete the expression of interest form.

I take care to review all expressions of interest thoroughly to ensure that the study is appropriate for all participants. Although I believe that all parents of children with congenital heart disease have meaningful and valuable experiences to share, the study must follow strict parameters regarding who is able to take part. This is because the method of analysis that will be used requires that all participants have similar shared experiences.

I have carefully reviewed your expression of interest form and, unfortunately, you are not eligible to take part in the study at this time because (insert reason). I understand that this may feel disappointing and encourage you to contact me if you have any concerns.

I would like to take this opportunity to thank you again for your interest in the research. I would also like to let you know that I would be happy to share a summary of the findings with you once the study is completed and assessed. Please let me know if this is something you would like to receive.

If you have any further questions or queries, please do not hesitate to contact me.

Kind regards,

Shannon Dandy
Trainee Clinical Psychologist
Lancaster University

**Appendix 4-G**
**Microsoft Teams guidance**

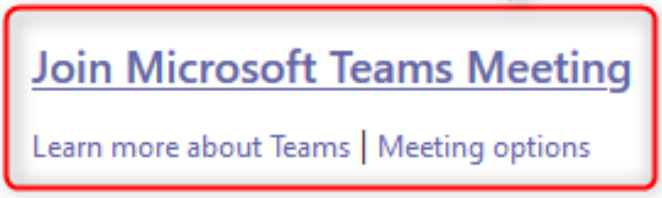
# Guidance for Microsoft Teams

Microsoft Teams is an online videoconferencing platform that allows remote video meetings to take place. The interview with Shannon will take place using Microsoft Teams, so below is some guidance on how to use it. It is recommended to use Microsoft Teams on a laptop or desktop device for the interview. If you have any further questions or queries, please contact Shannon at s.dandy@lancaster.ac.uk

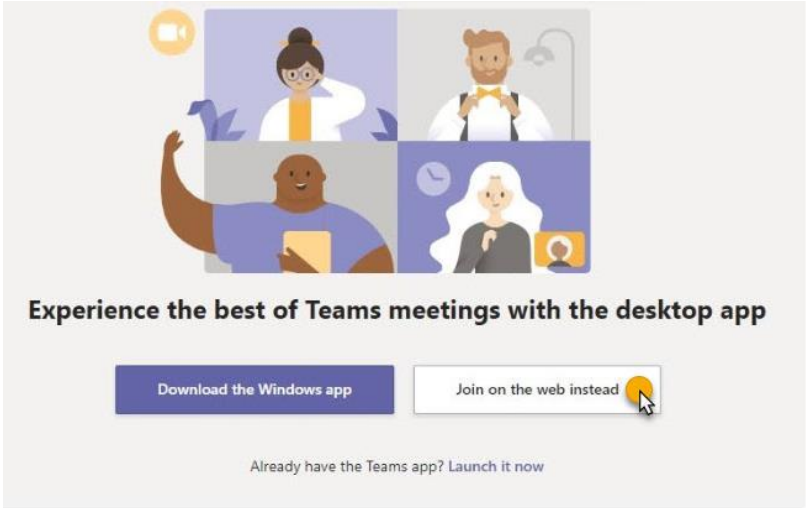
## Using a Laptop / Desktop Device (Online)

You can download the Microsoft Teams Desktop app or, if you don't want to download any software, you can choose to use the internet browser version instead. Below are some instructions on how to use the internet browser version of Microsoft Teams:

1. You will receive an email from Shannon confirming the agreed date and time of the interview. This will contain an invite to the Microsoft Teams Meeting. The link to join the meeting will look like this:



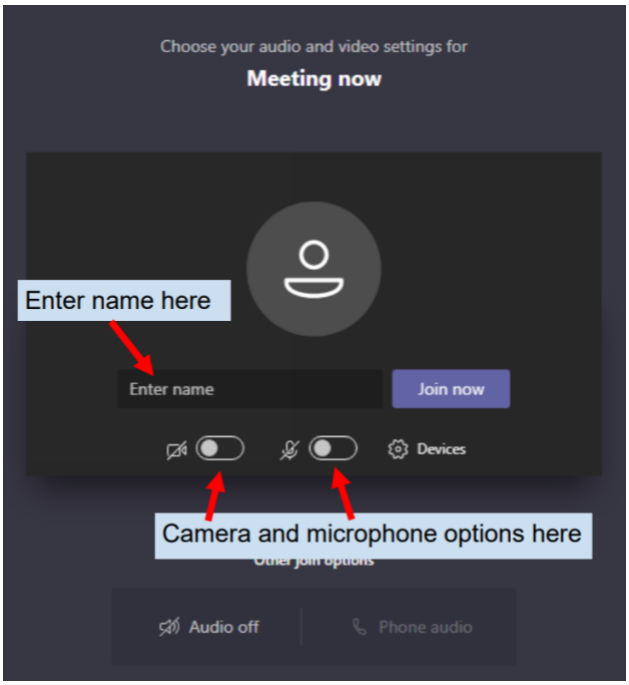

2. At the time of the interview, click on the link to the meeting. This will take you to a webpage that looks like this:




To access the internet browser version of Microsoft Teams (without downloading any software) click 'join on the web instead'.

3. The first time you do this, it will advise you that Microsoft Teams will use your microphone and camera. Click 'allow' to enable your microphone and camera. If you click block, you will not be able to attend the meeting.

4. The screen will then look like this:



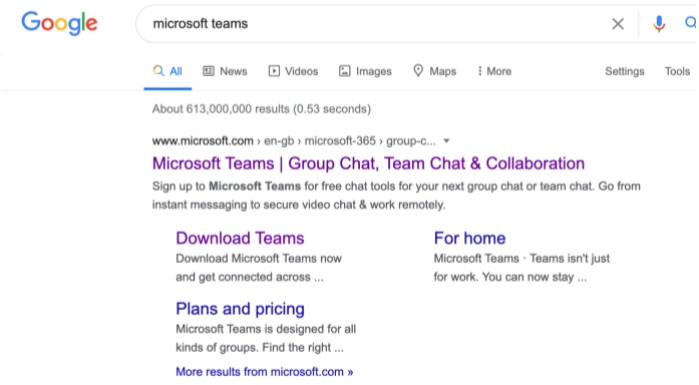

You can enter your name and adjust whether you want your camera and/or microphone to be turned on before joining the meeting.

5. Ensure you have a quiet, private space to talk. Once you are happy with your settings, click 'join now' to enter the meeting.

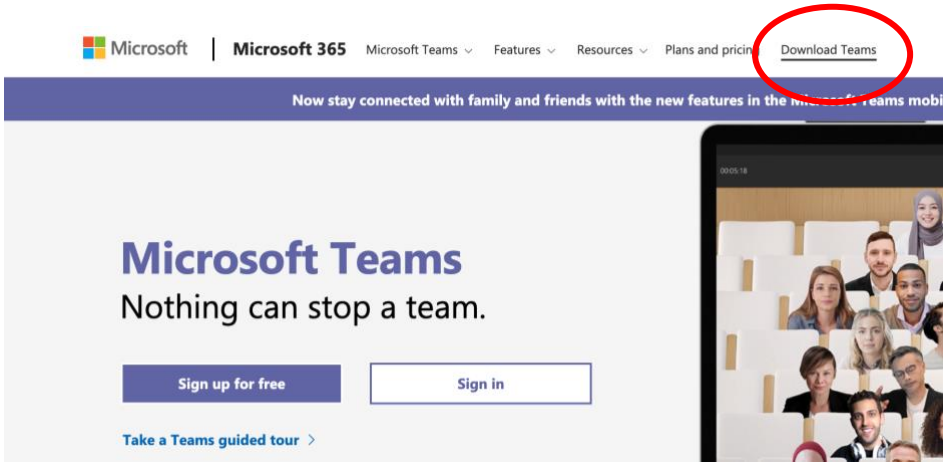
## Using Laptop / Desktop Device (Teams App)

Alternatively, you can download the free Microsoft Teams Desktop App. If you wish to download the app, **please do so in advance of the interview as software can sometimes take a while to install**. Instructions on how to download and use the Microsoft Teams desktop app are below:

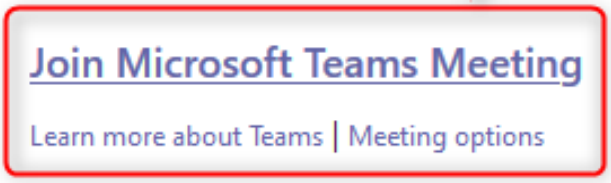
1. Search for 'Microsoft Teams' in your internet search engine and click on the link to the Microsoft website. It might look like this:



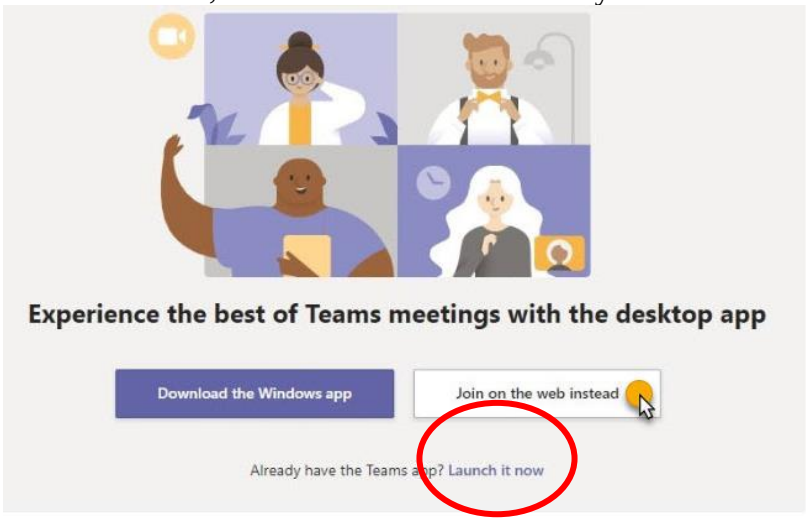
2. On the Microsoft website, click ‘download teams’:




3. Select download to desktop
4. This will open a pop out window where you can decide where to save the software on your laptop/desktop computer. Follow the download and installation instructions.
5. Once this is complete, you will be able to use the Microsoft Teams App. Shannon will have sent you an email confirming the agreed date and time of the interview as well as a link to the Teams meeting:

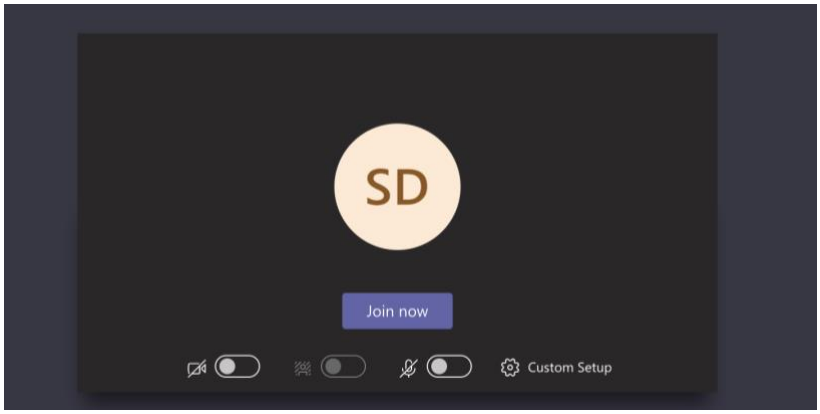



6. At the time of the interview, click on the link. It will take you to this screen:




You may be presented with a pop up that offers to take you straight to the Microsoft Teams app. You can choose to click on this or use the ‘launch it now’ link shown above.

7. The screen will look like this:

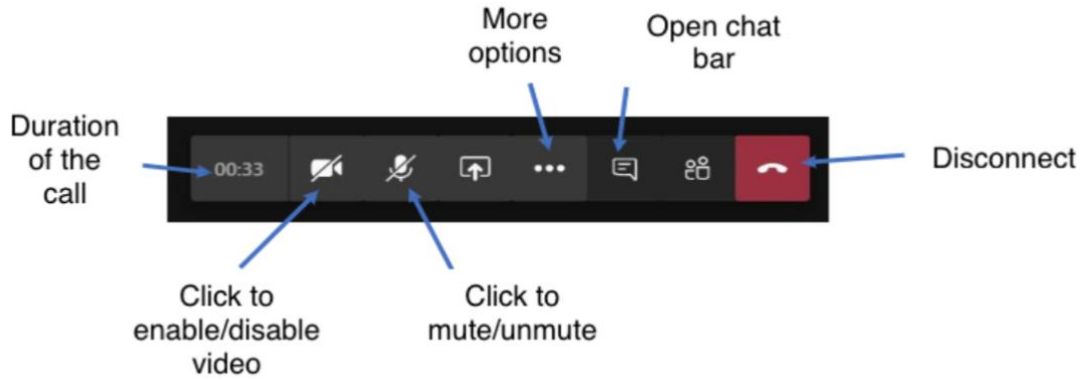



You can adjust whether you want your camera or microphone to be on.

8. Ensure you have a quiet, private space to complete the interview. Once you are happy with your settings, click 'join now' to enter the meeting.

## Features during the call

During the call there are several features you can use to turn on and off your audio or camera, use the chat function, or end the call. These are shown in an options bar towards the top of the screen, which looks like this:




At the end of the interview, you can use the red 'disconnect' button to leave the meeting.

**Appendix 4-H**
**Email response to participants who wish to be interviewed as a couple**

Dear (name),

Thank you very much for your interest in the research I am conducting regarding experiences of parents of children with single ventricle congenital heart disease. I really appreciate you taking the time to contact me.

From your correspondence I understand that, if you were to take part in the study, you would like to be interviewed alongside your (use the individual's wording, e.g. husband, wife, partner). Unfortunately, at the moment, the focus of the study is interviewing parents individually and therefore I am not completing interviews with couples at this time. The reason for this is to ensure that there is sufficient time within the interview to discuss each individual's experiences. However, I appreciate that this might feel disappointing.

It is possible that, in the future, this may change and that interviews with couples may become a part of the study. Would you be happy for me to retain your contact details and get in touch in the future, if this becomes the case? There is absolutely no obligation for you to agree to this or to agree to take part in future. Alternatively, if you would prefer that I did not contact you in the future please email me to let me know.

In addition, I would be happy to share a summary of the findings with you, regardless of whether you are able to take part or not, once the study is completed and assessed. Please let me know if this is something you would like to receive.

Finally, I would like to take the opportunity to thank you again for your interest in the research and wish you the best for the future. If you have any further questions or queries, please get in touch.

Kind regards,

Shannon Dandy
Trainee Clinical Psychologist
Lancaster University

**Appendix 4-I**
**Participant consent form**

**Study Title:** Having a child with single ventricle congenital heart disease: Parental experience of role and identity.

You have been invited to take part in this research exploring the experiences of parents of children with single ventricle congenital heart disease and, in particular, any impacts this may have on their own sense of role or identity.

Before you consent to participating in the study, please ensure that you have read the participant information sheet and have read each statement below. Please note that if you do not answer the telephone or video call at the time of the scheduled interview, the researcher will call again twice more after 5 and 10 minutes. If all calls go unanswered, the researcher will assume that you no longer wish to take part and will not persist in trying to contact you.

When you meet with the lead researcher, Shannon Dandy, she will ask you to state your name and the date. She will then read out each of the statements in turn and ask whether you agree to each one. This will take place before the interview begins and will be recorded. If you have any questions or queries before providing verbal consent at the start of the interview, please speak to Shannon as the lead researcher.

1. I understand that the process of verbal consent that is about to be completed will be audio recorded and this will be separate from any interview recording.

2. I understand that this audio recording of verbal consent will be stored for 10 years after the study has finished.

3. I confirm that I have read the information sheet and fully understand what is expected of me within this study.

4. I confirm that I have had the opportunity to ask any questions and to have them answered.

5. I understand that Microsoft Teams is the recommended medium for interviews and that if I have chosen to participate via other videoconferencing software or telephone the lead researcher is unable to guarantee anonymity of confidentiality for interviews due to the nature of these platforms.

6. I understand that my interview will be audio recorded and then made into an anonymised written transcript.

7. I understand that audio recordings of the interview will be kept until the recording has been transcribed and anonymised.

8. I understand that my participation is voluntary and that I am free to withdraw at any time before or during the interview without giving any reason. I understand that I can withdraw my interview data up to 2 weeks after the interview.

9. I understand that 2 weeks after the interview, my data will begin to be anonymised, transcribed, analysed and incorporated into themes. I understand that once this process begins, it will not be possible for my data to be withdrawn.

10. I understand that my anonymised interview transcript will be shared with the research supervisors, Dr Craig Murray and Dr Anja Wittkowski.

11. I understand that the information from my interview will be pooled with other participants' responses, anonymised and may be published; all reasonable steps will be taken to protect the anonymity of participants involved in this project.

12. I consent to information and quotations from my interview being used in reports, conferences and training events.

13. I understand that the researcher will discuss data with their supervisor as needed.

14. I understand that any information I give will remain confidential and anonymous unless it is thought that there is a risk of harm to myself or others, in which case the lead researcher (Shannon Dandy) will need to share this information with their research supervisor and any other relevant agencies.

15. I consent to Lancaster University keeping written transcriptions of the interview for 10 years after the study has finished.

16. I consent to take part in the above study.

**Appendix 4-J**
**Debriefing form**

**Study Title:** Having a child with single ventricle congenital heart disease: Parental experience of role and identity.

## Thank you for taking part in this study!

The study was exploring the experience of being the parent of a child with single ventricle congenital heart disease and any impacts this may have on parents' perceptions of their own role and identity.

**What happens next?**

Results from the study will be summarised and reported in a thesis paper that will be assessed as part of my Doctorate in Clinical Psychology qualification. The report may also be submitted to a peer-reviewed journal. If you have chosen to receive a summary of findings, these will be sent to you via email once the thesis has been assessed.

If you wish to withdraw your data, you can do so for up to two weeks after the interview by contacting me at s.dandy@lancaster.ac.uk. Please note that, after two weeks, the data will begin to be anonymised, transcribed, analysed and incorporated into themes. Therefore, it will not be possible to identify and withdraw your data.

**Further questions or complaints**

If you have any further questions or concerns relating to the study, please contact the lead researcher, Shannon Dandy, at s.dandy@lancaster.ac.uk or the research supervisor, Dr Craig Murray, at c.murray@lancaster.ac.uk.

If you have any concern or complaints regarding the study and do not wish to speak to the researcher, you can contact Dr Ian Smith (Research Director, Lancaster University Docotrate in Clinical Psychology) at i.smith@lancaster.ac.uk. If you would prefer to speak to someone outside of the Doctorate in Clinical Psychology Programme, you may also contact Dr Laura Machin at l.machine@lancaster.ac.uk.

**Sources of Support**

You may wish to seek further support or resources after taking part in the study. If you experience distress either as a result of taking part, or in the future, please contact your GP. There are also alternative sources of support or information listed below:

- Your child's Cardiology team (sometimes there is also a Clinical Psychologist who works within the Cardiology team who may be able to offer support).
- Little Hearts Matter: A national charity supporting children with single ventricle congenital heart disease and their families:
    - 0121 455 8982
    - Lhm.org.uk
- Children's Heart Federation: A national charity for children with heart conditions and their families
    - 0300 561 0065
    - Chfed.org.uk

- Other sources of emotional support may also be helpful, for example the Samaritan's Helpline: 116 123

**Finally, thank you once more for taking part in the study and best wishes for the future.**

**Appendix 4-K**
**Example semi-structured interview topic guide**

**Key**
Blue text – pre-interview procedural discussion points and questions
Black text – interview topic guide & questions
Red text – post-interview procedural discussion points and questions

| Main topic | Examples of sub-topics & questions | Examples of further prompts |
|---|---|---|
| Introductions & demographics | • Welcome to the interview, confirm participant is in a quiet & comfortable place<br>• Introductions | • My name and title, what a trainee clinical psychologist is, why I am doing the research |
| | • Talk through participant information sheet, check inclusion / exclusion criteria | • Confirm eligibility criteria and demographics<br>• Child's age (16 years or under) condition & completion of Fontan surgery<br>• Surgery must have been at least 6 months ago<br>• Must reside in the UK<br>• Child must not currently be in hospital, have undergone transplant or passed away |
| | • Obtain & **record** verbal consent<br>• Obtain preferred pseudonym | • Once obtained consent, stop recording<br>• Reference confidentiality & keeping information private to introduce purpose of pseudonym.<br>• What name would you like to have on the interview transcript and any quotes I use from our conversation? |

**Start new audio recording**

| | | |
|---|---|---|
| Re-orient to purpose of interview & introduce to interview process | • I am interested in how you might make sense of your parental role and identity (how you see yourself) in relation to having a child with single ventricle CHD. I would like to hear about your perceptions of this along your journey as a parent. Today we will have approximately one hour to talk. I may say some things or ask some questions, but I will try to keep this to a minimum. | |
| Condition trajectory | • Could you tell me a bit about your child and their condition?<br>• What were some of the key events along this journey? | • What was it like when you received the diagnosis? |
| Parenting & parental role | • Is your child with CHD your first child?<br>• When did you find out about your child having single ventricle CHD?<br>• How did you understand / make sense of that? | • If no, ask briefly about other children. |
| | • How would you describe your role as a parent of a child with single ventricle CHD? | • E.g., in relation to child, others & professionals. |
| | Expectations of parenthood:<br>• How does this compare to what you were expecting? | • E.g., pre-diagnosis<br>• (If relevant) How does this compare to your role when parenting your other child(ren)? |
| | Changes to parental role<br>• Have you noticed any changes in your role at any time throughout your journey with your child? | • E.g., at diagnosis, child being in hospital, ICU, surgery, transition home from hospital.<br>• How have you coped / adapted to this?<br>• What has been easiest / hardest? |

| | | |
|---|---|---|
| | • How has having this child changed your parenting style (from previous children or from expectations) | • E.g., stricter / more lenient |
| | • What have you noticed about your parental role as your child has gotten older? | • E.g., transition to school |
| Impact on identity | • How has having a child with single ventricle CHD impacted upon how you see yourself?<br>• How has having a child with single ventricle CHD impacted on other aspects of your identity (i.e., as a partner, friend, employee) or your relationship with them? | |
| | • Have other aspects of your identity receded since having a child with single ventricle CHD?<br>• To what extent has the amount of pleasure you take from these other identities been affected by having a child with single ventricle CHD? | • If no, how have you balanced these identities and associated commitments? |
| | • How has having a child with single ventricle CHD impacted how others see you? | |

**Generic Prompts**

- Can you tell me more about that?
- Could you describe that for me?
- Could you give an example of that?
- What happened next?
- What was that like?
- How did that feel?
- How did you cope?

| | | |
|---|---|---|
| Debrief & closing questions | • Express thanks for taking part<br>• Acknowledge it can be difficult to share experiences of having a child with CHD<br>• Allow time for any questions<br>• Explain that debriefing document will be sent to email<br>• Ask whether participants would like to receive a summary of results once the thesis has been assessed | • How did you find the interview?<br>• How do you feel now? |

Applicant: Shannon Dandy
Supervisor: Dr Craig Murray
Department: Division of Health Research (Clinical Psychology) FHMREC Reference: FHMREC20023

14 January 2021

**Re: FHMREC20023**
**Having a child with single ventricle congenital heart disease: Parental experience of role and identity**

Dear Shannon,

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

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;

- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);

- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information. Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

Annie Beauchamp,
Research Ethics Officer, Secretary to FHMREC.

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

1. Name of applicant:

Shannon Dandy

2. E-mail address and phone number of applicant:

s.dandy@lancaster.ac.uk
07411135890

3. Title of project:

Having a child with single ventricle congenital heart disease: Parental experience of role and identity

4. FHMREC project reference number:

FHMREC20023

5. Date of original project approval as indicated on the official approval letter (month/year):

14/01/2021

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

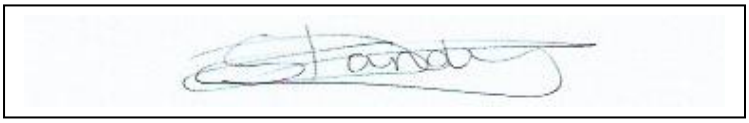
The original application specified recruitment would take place via the social media and email lists of children's heart charities. An amendment is being submitted to request approval for the lead researcher to also create a research Twitter account. The Twitter account would be used for the sole purpose of advertising the study using the materials already approved: the study advertisement (appendix B) and a link to the participant information sheet (appendix A). Active online charities and groups for parents of children with CHD in the UK will be contacted to ask to consider 're-tweeting' the post. Wording would state: Are you the parent of a child with single ventricle congenital heart disease? Opportunity to be involved in research here: (link to participant information sheet).

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

The amendment is being requested to provide an extra method of advertising the study, in order to maximise accessibility to the study and opportunities for participant recruitment.

Guidance:

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



23/02/2021

Applicant electronic signature: Date

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

**Project Supervisor name** (if applicable): Date application discussed

Dr Craig Murray

23/02/2021

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

Applicant: Shannon Dandy
Supervisor: Craig Murray, Anja Wittkowski
Department: DHR
FHMREC Reference: FHMREC20114 (amendment to FHMREC20023)

02 March 2021

**Re: FHMREC20014 (amendment to FHMREC20023)**
**Having a child with single ventricle congenital heart disease: Parental experience of role and identity**

Dear Shannon,

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

As principal investigator your responsibilities include:

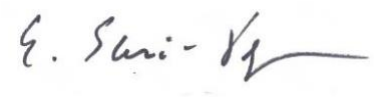
- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;

- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g., unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);

- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information. Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,



Dr. Elisabeth Suri-Payer
Research Ethics Officer, Secretary to FHMREC

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

1. Name of applicant:

   Shannon Dandy

2. E-mail address and phone number of applicant:

   s.dandy@lancaster.ac.uk
   07411135890

3. Title of project:

   Having a child with single ventricle congenital heart disease: Parental experience of role and identity

4. FHMREC project reference number:

   FHMREC20023

5. Date of original project approval as indicated on the official approval letter (month/year):

   14/01/2021

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

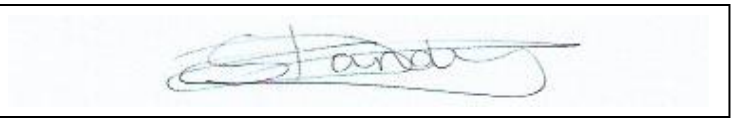
   The original application and a subsequent amendment specifies that recruitment takes place via social media and email lists of children's heart charities and a dedicated Twitter account. An amendment is being submitted to request approval for further advertisement on wider social media platforms (e.g., Facebook, Reddit) using the same advertising materials already approved (appendix B).

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

   The amendment is being requested to provide an extra method of advertising the study, in order to maximise accessibility to the study and opportunities for participant recruitment.

Guidance:

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



16/06/2021

Applicant electronic signature: Date

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

**Project Supervisor name** (if applicable): Date application discussed

Dr Craig Murray

17/06/2021

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

Applicant: Shannon Dandy
Supervisor: Craig Murray, Anja Wittkowski
Department: DHR
FHMREC Reference: FHMREC20155 (Amendment FHMREC20023/114)

02 July 2021

**Re: FHMREC20155 (Amendment FHMREC20023/114)**
**Having a child with single ventricle congenital heart disease: Parental experience of role and identity**

Dear Shannon,

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

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;

- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g. unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);

- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information. Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

T. Morley

Tom Morley,
Research Ethics Officer, Secretary to FHMREC.

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

1. Name of applicant:

   Shannon Dandy

2. E-mail address and phone number of applicant:

   s.dandy@lancaster.ac.uk
   07411135890

3. Title of project:

   Having a child with single ventricle congenital heart disease: Parental experience of role and identity

4. FHMREC project reference number:

   FHMREC20023

5. Date of original project approval as indicated on the official approval letter (month/year):

   14/01/2021

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

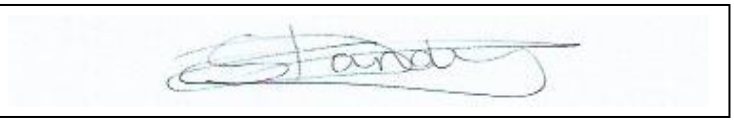
   The original application states that an inclusion criterion for participants is that their child with single ventricle congenital heart disease is aged 16 years or younger at the time of interview. The rationale for this criteria was to promote homogeneity among participants and reduce the burden on memories being recalled. Due to difficulties with recruitment, the amendment proposes to change this criteria to allow participants with children aged 25 years and under to take part, to expand the potential sample pool. The lead researcher has already received expressions of interest from individuals who would meet the new criteria.

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

   The amendment is being requested due to ongoing difficulties with recruitment of participants that meet all inclusion criteria, in the context of only a small number of children having this condition.

Guidance:

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



19/07/2021

Applicant electronic signature: Date

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

**Project Supervisor name** (if applicable): Date application discussed

Dr Craig Murray

19/07/2021

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

Applicant: Shannon Dandy
Supervisor: Dr Craig Murray
Department: DHR
FHMREC Reference: FHMREC20182 (amendment to FHMREC20023)

27 July 2021

**Re: FHMREC20182 (amendment to FHMREC20023)**
**Having a child with single ventricle congenital heart disease: Parental experience of role and identity**

Dear Shannon,

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

As principal investigator your responsibilities include:

- ensuring that (where applicable) all the necessary legal and regulatory requirements in order to conduct the research are met, and the necessary licenses and approvals have been obtained;

- reporting any ethics-related issues that occur during the course of the research or arising from the research to the Research Ethics Officer at the email address below (e.g., unforeseen ethical issues, complaints about the conduct of the research, adverse reactions such as extreme distress);

- submitting details of proposed substantive amendments to the protocol to the Research Ethics Officer for approval.

Please contact me if you have any queries or require further information. Email: fhmresearchsupport@lancaster.ac.uk

Yours sincerely,

T. Morley

Tom Morley,
Research Ethics Officer, Secretary to FHMREC.