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An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

And Clinical Research Portfolio

Evelyn Marscheider,

BSc (Hons), MSc

Submitted in partial fulfilment of the requirements for the degree of Doctorate
in Clinical Psychology

Institute of Health and Wellbeing
College of Medical, Veterinary and Life Sciences
University of Glasgow

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Chapter One: Systematic Review

Experiences of transition from paediatric to adult congenital heart disease care:
a synthesis of the literature

Evelyn Marscheider
BSc (Hons), MSc

Submitted in partial fulfilment of the requirements for the degree of Doctorate
in Clinical Psychology

Address for correspondence:

Evelyn Marscheider / Dr Naomi White

Institute of Health and Wellbeing

College of Medical, Veterinary and Life Sciences

University of Glasgow

Administration Building, Gartnavel Royal Hospital

1055 Great Western Road, Glasgow, G12 0XH

Email: e.marscheider.1@research.gla.ac.uk, naomi.white@glasgow.ac.uk

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Abstract

Background

Patients with Congenital Heart Disease (CHD) are living longer, healthier lives. Thus, improved transitions from paediatric to adult care are required. Poor transition can lead to increased morbidity, psychological distress and financial expense. A greater understanding of patients' experiences is needed to inform transition interventions, improve positive outcomes, and reduce lapses in care.

Aim

The current review aims to synthesise the qualitative research exploring patients' experiences of transition from paediatric to adult CHD care.

Method

An electronic database search was performed. The search was limited to peer-reviewed studies published in English after January 2001. Analytical themes were developed through thematic synthesis of 10 studies.

Results

One overarching theme was identified: The importance of being an active participant in individualised care. Three further subthemes were developed: "I was given a choice and I liked that" - opportunities for involvement; "It was like they had to tell me everything, it was horrible!" - individualised opportunities; and "Take in the whole person" - the need for holistic care.

Discussion

Participants valued active involvement in transition at a pace individualised to wants and developmental needs thus highlighting the importance of patient-centred care during transition. Transition programmes should be adequately flexible to attend to individual differences to increase engagement, reduce lapses in care and reduce negative associated outcomes.

Key words: congenital heart disease, transition to adult care, experiences

Background

Historically, most Congenital Heart Disease (CHD) patients did not survive into adulthood. With medical advances, patients are surviving longer, with healthier lives into adulthood. The focus has therefore shifted from survival to ensuring successful transition into and care throughout adulthood. Poor transition can lead to negative outcomes including increased morbidity, emotional distress, and financial expense. These outcomes can occur as a result of poor transition, leading to lapses in care. Although there is a need for a better operational definition (Heery et al., 2015), a lapse of care generally refers to a substantial gap between leaving paediatric CHD care and initiating specialised adult CHD care. Yeung and colleagues (2008) found that patients who presented after a lapse in care were more likely to require urgent cardiac interventions and have untreated cardiac-related conditions. Participants frequently reported that they did not continue to seek care due to insufficient knowledge regarding the importance of continued care and monitoring, highlighting a potential barrier in education or communication during the transition process.

As prompted by the introduction of guidelines for best practice in transitioning young people with chronic illness (Forbes et al., 2001; Foster et al., 2001) and the development of national and international standards (NHS England, 2016; Sable et al., 2011; Scottish Congenital Cardiac Service, 2018; Warnes et al., 2008), cardiology services have developed programmes to ensure the continuity of high-quality care. Such models have been reported to show varying outcomes (Lee et al., 2017).

Transition Experiences

For many patients with CHD, transition to adult care can present with multiple challenges and barriers (Lee et al., 2017). Heery and colleagues (2015) conducted a systematic review examining experiences of transition among young people with CHD. They found high rates of lapses in care for young people after leaving paediatric cardiology. Data highlighted that young people had

insufficient knowledge about their condition to facilitate adequate transition. They concluded that young people looked for continuity of person-centred care, youth-orientated facilities, and for parents to be involved.

Rationale for the Current Review

Qualitative synthesis of studies focusing on patients' experiences of transition from paediatric to adult CHD care should inform the development of interventions to reduce poor transition and associated negative outcomes. Building on the findings of Heery and colleagues (2015), this review aimed to provide an updated synthesis and quality appraisal of the literature. Heery and colleagues review found no research specifically on experiences of structured transition programmes nor the experiences of patients' post-transfer. In addition, to increase sensitivity, studies that included other chronic illnesses were not excluded providing CHD-specific data were extractable.

The main aims of this review were to appraise the quality and synthesise findings from qualitative research of patients' experiences of transition to adult CHD care.

Method

Search Strategy

A systematic search was conducted of the following databases:

- Medline (Ovid)
- EMBASE (Ovid)
- PsycINFO (EBSCO)
- CINAHL (EBSCO)
- Psychology & Behavioural Sciences (EBSCO)
- Web of Science Core Collection (Web of Science)

The search strategy combined three series of terms which defined the congenital heart condition, transition process and qualitative methodology. Search terms were mapped to subject headings within databases. An example of the full search conducted in Medline is detailed in appendix 1.2. The following search terms were used:

1. Congenital heart OR heart defect* OR heart syndrome OR congenital abnormalities or cardiovascular abnormalities
2. Transition* OR transfer* OR continuity* OR move* OR hand-over* OR retention
3. Qualitative OR interview* OR experience*

Inclusion / Exclusion Criteria

All qualitative designs were eligible for inclusion and papers were not excluded on the basis of quality assessment. The search was limited to peer-reviewed studies published in English between January 2001 onwards, due to the publication of guidelines for best practice in transitioning young people with chronic illness (Forbes et al., 2001; Foster et al., 2001).

Results of Search

The search strategy identified 6948 citations, including 4861 unique citations, but 4825 were removed through screening (see Figure 1). The full text of 36

papers were reviewed, 10 of which met the inclusion criteria. Hand searches of references and grey literature did not identify any additional eligible papers.

Data Synthesis

A data extraction tool (appendix 1.3) was used to record only findings relevant to the review aims. Despite the acknowledgement of the value of qualitative research for health care, the ‘gold standard’ method of qualitative synthesis is an area of debate and evolution. Noyes and colleagues (2008) state that the choice of method depends on the type and scope of the review, the pool of available evidence, the expertise of the researchers, and the available resources. On these grounds, thematic synthesis was adopted, allowing conclusions to be drawn across heterogeneous methodologies. It also analyses characteristics of the studies to explore whether these help explain differences in perspectives that arise (Barnett-Page & Thomas, 2009), relevant when considering varying international healthcare contexts. Thomas and Harden’s (2008) steps of thematic synthesis were followed (see appendix 1.4). An example of an analytical theme and illustrations can be found in appendix 1.5.

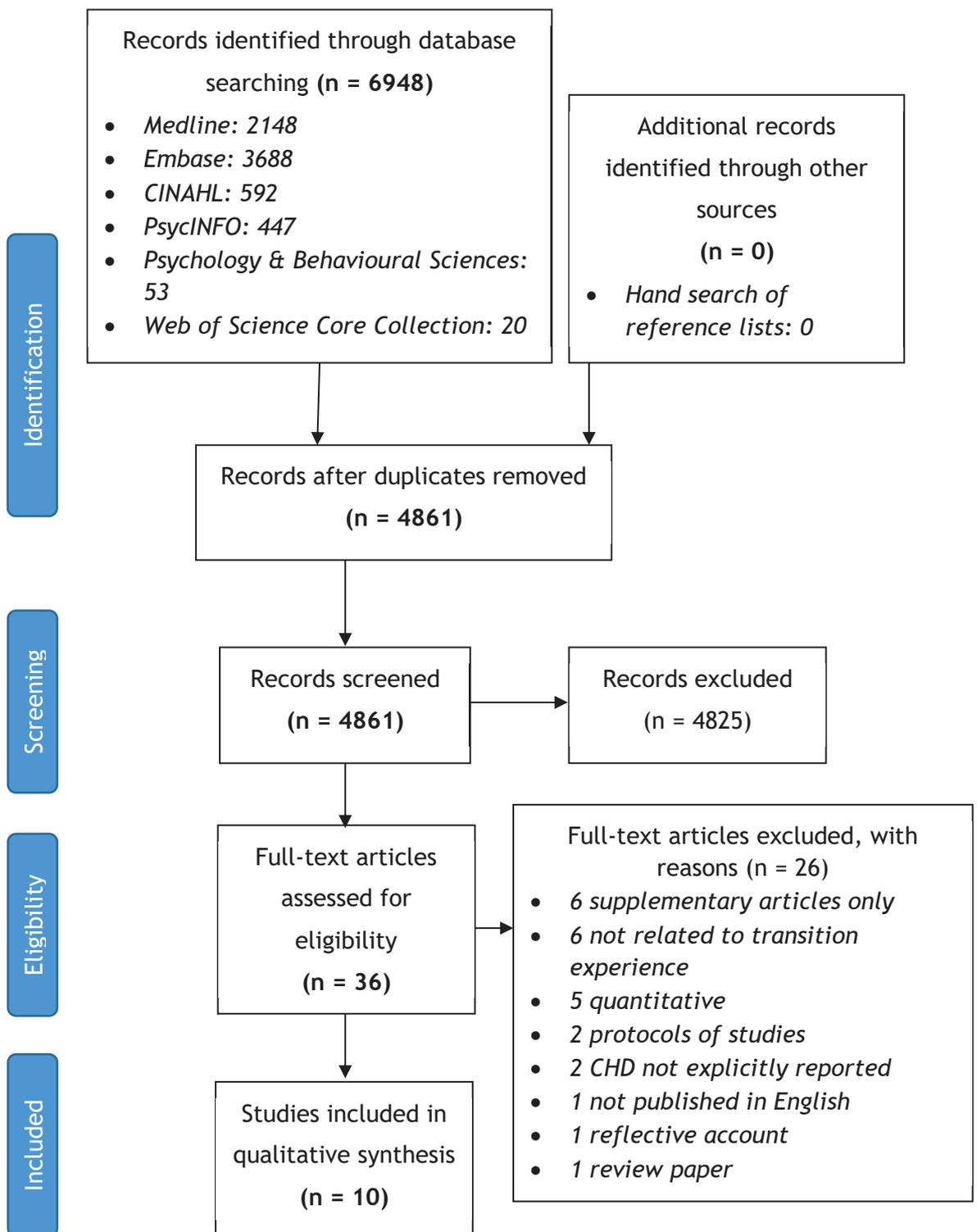


Figure 1. PRISMA Flow Diagram - Adapted from Moher, Liberati, Tetzlaff, & Altman (2009)

Quality Appraisal

Quality appraisal facilitates understanding of the transferability of findings (Whiting et al., 2017). Over 100 appraisal tools for qualitative research have been developed, highlighting the lack of consensus over quality criteria. The 10-item Joanna Briggs Institute (JBI) Critical Appraisal Checklist for Qualitative Research (Joanna Briggs Institute, 2016) was considered to be consistent with the resources, aims and assumptions of this synthesis including its emphasis of researcher reflexivity, application to all qualitative methodologies, and its development from evidence base (Majid & Vanstone, 2018).

The author used the JBI Checklist to assess qualitative indicators of studies' strengths and weaknesses; which were considered during analysis. 3 studies were selected randomly to be co-assessed. Disagreements in quality assessments were discussed and recorded in a reflective log. Final comparisons were adequately similar with 96.7% agreement across the 10 items for the 3 studies; therefore it was deemed unnecessary to co-rate additional studies.

Results

An overview of the 10 included studies is given in Table 1. The studies originated from 6 countries. Data were collected using face-to-face interviews (n=6), telephone interviews (n=2), focus groups (n=1) and postal surveys (n=1). Data collected perspectives from adolescents pre-transfer (n=3), adolescents during transfer (n=1), young adults post-transfer (n=2), adults post-transfer (n=1), and both adolescents and adults pre- and post-transfer (n=3). Sample sizes ranged from 10-40 participants.

The findings are presented under one overarching theme: The importance of being an active participant in individualised care. Participants wanted to be actively involved in their transition but at a pace that was individualised to their wants and developmental needs. They also wanted to be considered as a whole person, with interest shown beyond their heart condition. This in turn facilitated their personal connections and trust with HCPs. Three subthemes were developed to illustrate these findings: “I was given a choice and I liked that” - opportunities for involvement; “It was like they had to tell me everything, it was horrible!” - individualised opportunities; and “Take in the whole person” - the need for holistic care. A breakdown of themes is included in appendix 1.6.

Table 1

<i>Studies included in systematic review</i>		<i>Method of analysis</i>		
<i>First author, year, country</i>	<i>Characteristics of patients</i>	<i>Transition process</i>	<i>Data collection</i>	<i>Findings</i>
Coyne, 2019 Ireland	14-25 years n = 16 with CHD	No transition clinics Referral letter	Semi-structured interviews	<ol style="list-style-type: none"> 1. Transition process and preparation: <ol style="list-style-type: none"> a. Transition practices & preparation b. Timing 2. Expectations of adult services: <ol style="list-style-type: none"> a. Acceptance & loss b. Culturally different environment c. Acceptance of move d. Loss of security & relationships e. Concerns over shift in responsibility 3. Transition to adult services - a culture shock <ol style="list-style-type: none"> a. No man's land b. Culture shock c. Shift in responsibility 4. Flourishing or floundering in new roles
Catena, 2018 Canada	18-25 years n = 21 Moderate or complex CHD n = 13 transferred n = 8 not yet transferred	Toronto: Good 2 go programme Edmonton: No transition initiatives Article removed locations for	Structure telephone interviews	<ol style="list-style-type: none"> 1. Perspectives on paediatric settings and relationships: <ol style="list-style-type: none"> a. The paed cardiology setting "A little home" b. Paed cardiology healthcare team "A personal connection, a comfort thing" 2. Perspectives on the parents' role <ol style="list-style-type: none"> a. Parental involvement: Letting go b. Young adults holding on "She knows more about it than me" 3. Perspectives on transfer

Keir, 2018 Canada	38-49 years n = 10 Complex CHD Childhood surgery	Not reported	Interviews	Narrative analysis	<p>anonymity.</p> <p>a. Negative experiences “Pushed” and “Shoved” b. Positive experiences “Not such a kid thing anymore”</p> <p>‘Transition to adult care’</p> <ul style="list-style-type: none"> - Insufficiently education and preparation for future - Misperceptions of limitations - Challenges participating in adult health care decisions
Burström, 2017 Sweden	14 - 18 years n = 17 Moderate to complex CHD	Unstructured transition preparations - no set guidelines	Four focus groups (3-5 in each group)	Content analysis	<p>Becoming a manager of the condition</p> <ol style="list-style-type: none"> 1. Sufficient knowledge about health 2. Be a participant in the care 3. Parental support 4. Communicating with others about health <p>Age differentiated discussions: - older: Transition planning, information and transfer - younger: Communication frustrations & disease management</p>
Burström, 2016 Sweden	16 - 18 years n = 13 Moderate to complex CHD	During transition Structure of transition not reported	Semi-structured interviews	Content analysis	<ol style="list-style-type: none"> 1. Change of relationships - natural step indicating change in relationships; continuity major concern 2. Knowledge and information - perception, understanding & timing of information; parents play crucial role 3. Daily living - parents central for support; frustrations of physical limitations; environmental restrictions
Lopez, 2015 USA	Adolescents: 16-20 years n = 20 Adults: 21-40 years n = 20	Varied transition experiences	Semi-structured telephone interviews	No specific method quoted - data qualitatively coded with themes identified	<p>Adolescent:</p> <ol style="list-style-type: none"> 1. Medical knowledge and concerns: 50% wished more information; 50% concerns about being an adult 2. Preferred methods of information: 85% direct conversation 3. Mentorship: 60% helpful to talk to CHD adult

	Moderate or severe CHD					
Asp, 2015 Sweden	20 - 24 years n = 16 CHD surgery Moderate to complex CHD	Varied preparation for transfer	Semi-structured interviews	Content analysis	<p>4. Social media: 90% interest in contact with CHD adolescent</p> <p>5. Checklist: 55% internet-based transition checklist helpful</p> <p>Adult:</p> <ol style="list-style-type: none"> 1. Medical knowledge: 45% wished more CHD information 2. Information received/not received as adolescents: 40% more information about exercise, nutrition & lifestyle 3. Self-advocacy, pregnancy, dental and mental health: importance of topics in adolescence 4. Preferred methods of information: talking to a doctor 5. Checklist: 80% saw benefit 	
Moola, 2011 Canada	11 - 17 years Complex CHD n = 50 (CHD n = ?)	Not reported	Semi-structured interviews	Thematic analysis	<ol style="list-style-type: none"> 1. Feeling secure during the transfer process 2. Experiencing trust in the care 3. Expecting to be involved 4. Assuming responsibility for one's health is a process 5. Lack of knowledge leads to uncertainty <p>The future as uncertain terrain:</p> <ul style="list-style-type: none"> - deteriorating health - occupational restrictions - motherhood 	
Moons, 2009 Belgium	15 - 17 years n = 14	No formal transition program	Semi-structured interviews	Phenomenologic (Colaizzi)	<ol style="list-style-type: none"> 1. Leaving paediatric cardiology was viewed as normal 2. Leaving behind familiar surrounding 3. Positive wait-and-see attitude toward the ACHD program 4. Adjusting to a new environment 5. Need for better information 6. Shift in roles between the adolescent and his parents 	

Wray, 2008 UK	17 - 20 years n = 38	No formal transition program	Postal survey	Thematic analysis	<ol style="list-style-type: none"> 1. Involvement of carers <ol style="list-style-type: none"> a. have family/support network around 2. Communication <ol style="list-style-type: none"> a. being informed, bring prepared 3. Care processes/caring interactions <ol style="list-style-type: none"> a. being involved in decision making - to have 'a voice' or 'more of a say' b. have the same standard of care - reassurance of specialist expertise c. gain confidence in new doctors - get to know new team quickly d. to 'fit in' with new team e. not feel lost in adult care f. records kept safe
<p><i>Notes. Abb = HCP: health care professionals; paed: paediatrics; ACHD: adult congenital heart disease; exp.: experiences; PCC: paediatric cardiac clinic</i></p>					

“I was given a choice and I liked that” - opportunities for participation

Participants across 8 studies emphasised the importance of being actively involved in healthcare decisions. This included being heard with HCPs seeking and incorporating their perspectives, opinions and choices in care plans. For some, transition facilitated a positive increase in participation in their care; which facilitated an increase in engagement, knowledge and sense of responsibility for their condition.

“I was given a choice and I liked that because it made me feel that I was getting older and was given more responsibility.” (Asp et al., 2015, p6).

“The most important thing to me when transferring to the adult clinic was that all the decisions were made with me involved and I was able to talk about what was going to happen when I saw the doctors, which I never had a chance to do in the children’s clinic.” (Wray et al., 2008, p571).

“I want to know everything that there is to know about my heart condition [...] cause I was so young when I had surgery to know what was going on, but now that I am older, I want to understand what is going on with me, and to understand what might happen in the future with me.” (Moola, 2011, p845)

Post-transfer young adults described that insufficient education opportunities during adolescence became a barrier to participation in adult healthcare.

It’s hard to take ownership because I didn’t know what my condition was. (Keir et al., 2018, p745).

For others, a lack of involvement and preparation appeared to shape negative experiences during transition and transfer. They described feeling “shoved right into it” (Catena et al., 2018, p596), with transfer often coming as a surprise. The desire to be included in transition over a prolonged period of time was

expressed. Some adolescents desired this information and involvement two or three years ahead. A lack of participation caused uncertainty and insecurity.

“I think it was just the last appointment at outpatients. It was thanks very much, ‘bye, bye, best of luck’. It’s just the way it was done. There was no ‘come in and we’ll have a chat about this and what do you think and how would you feel.’” (Coyne et al., 2019, p4067).

“He [paediatric cardiologist] suddenly said, this is your last check-up in the paediatric cardiac unit and they hadn’t said anything about it before. So it was very sudden.” (Asp et al., 2015, p5).

“My only gripe is that I didn’t know what was going to happen in advance and I did find that a bit awkward [...] it was all a bit unexpected.” (Moons et al., 2009, p319).

Opportunities for involvement in their care were also influenced by parents. During paediatric care, parents took the central role whereas transition was a time when adolescents sought increased autonomy. While some valued ongoing parental contributions for emotional support and a sense of safety during transition, others expressed frustrations for overly involved parents.

“My mother just took control. She’s like that ‘cause I’m the youngest, and the heart condition, I don’t like that.” (Catena et al., 2018, p595).

“She says I’m forgetful, so she wants to come and hear stuff from the doctors herself. But I keep telling her that she had to let me go sometimes just by myself.” (Catena et al., 2018 p595).

**“It was like they had to tell me everything, it was horrible!” -
individualised opportunities**

Although the importance of opportunities for involvement was expressed, it was also considered important across all studies that individual differences were considered. This particularly related to tailoring education around developmentally appropriate topics that were important to adolescents. These included alcohol use, exercise and nutrition, birth control and the risks associated with pregnancy and childbirth. Adults post-transfer reflected on the need for more information about mental health issues.

“Addressing concerns that nobody ever asked me before; like what does it mean that I have a heart condition when I wanted to have babies?” (Catena et al., 2018, p596).

“I think it would be beneficial when you’re transitioning and trying to learn how to cope and all of that and then differences between actual chest pain and anxiety.” (Lopez et al., 2015, p567).

In addition, adolescents highlighted the need for this information to be tailored so that the information did not appear exaggerated and intrusive.

“I must say, about the consequences, oh my god how they spoke about drinking and sex at my last visit. If you drink you’re going to die or end up in an emergency room. You are not allowed to get drunk and if you have unprotected sex you’re going to die. Oh my god. It was like they had to tell me everything, it was horrible! [...] You have to individualise such info.” (Burström et al., 2017, p884).

When information was not individualised to cognitive ability, the chances of misunderstanding and misinterpretations increased. Information was often described as given too fast with the excessive use of medical terminology and

jargon. On one occasion, it was described how the lack of tailored delivery led to the young adult seeking a transfer of physician.

“Like that time when I was in the adult clinic and they talk over my head, believing I understood what they were talking about [...] When they talk to me using a lot of medical terms it’s too much for me to take in.” (Asp et al., 2015, p7).

When information was delivered at developmentally inappropriate times in a way that the adolescent did not understand or was uninterested to learn, this often had a negative impact on education as they were reluctant to know more about their condition or not ready to process the information.

“When I was younger, I kind of just listened and agreed with whatever they said.” (Catena et al., 2018, p596).

Often it was reported that adolescents lacked opportunities for individualised education specific to their condition, leading to insufficient knowledge of the long-term impact including signs and symptoms of deterioration. This was highlighted as problematic due to the variability of CHD diagnoses.

“I wish I would have had a better understanding specifically of what I could and could not do, as far as working out, playing sports and stuff like that [...] without resulting in causing more problems to my situations.” (Lopez et al., 2015, p566).

“Should there be any complications with my heart again, as they don’t know if the heart will last for ten years or the rest of my life, or another two months, it would have been rather ‘fun’ to know the symptoms, what they are, what happens in order to be able to detect in time if something is not quite as it should be.” (Asp et al., 2015, p7).

Individual differences could also be seen in the preferred methods for delivering information. While some preferred to receive all information from HCPs, others would have like the opportunity to learn information from peers or an adult who had been through similar CHD related situations.

“Maybe talking to them could give me a different or a better perspective on certain things like how to take care of myself.” (Lopez et al., 2015, p565)

“Take in the whole person” - the need for holistic care

A shared theme across participants in 6 studies highlighted the importance of holistic care where professionals showed interest in other issues besides their heart condition. This helped participants to develop personal connections with HCPs and made them feel safe and cared for. It was considered important that the staff recognised them, addressed them by their first name and were available if they had any questions.

“It’s of great importance to me that the doctors don’t just examine my heart but take in the whole person.” (Moons et al., 2009, p319).

For those who had not transferred to ACHD the importance of holistic care was reflected in negative expectations that they would be treated as “another number” (Catena et al., 2018, p595) in ACHD services.

“I think it’s going to be a different atmosphere, like I think it’s going to be very kind of ‘just get the job done’.” (Coyne et al., 2019, p4067).

For those who had transferred, personal connections and holistic care, that had been fondly received in paediatric care, were often described as missing from initial interactions with ACHD providers.

“In the kid’s hospital everybody cared a little bit more, they’d try to make your stay comfortable and happy. In the adult hospital everyone’s just so busy, you don’t really get to have those relationships with your, the caregivers.” (Catena et al., 2018, p595).

“Feel vulnerable because there doesn’t seem to be the same effort made by staff to get to know you as there was in paediatrics.” (Wray et al., 2008, p572).

However, it was also reflected at post-transfer that the adjustment to and the building of new relationships would take time.

“It’s the type of thing where over the years hopefully [they’ll] be able to gain a little bit more of that personal one-on-one attitude.” (Catena et al., 2018, p595).

Quality Appraisal

The JBI Critical Appraisal Checklist for Qualitative Research (Joanna Briggs Institute, 2016) was used to identify strengths and weaknesses of the included studies. Due to the lack of consensus around assessing the quality of qualitative studies, no studies were excluded based on quality. In terms of strengths, all studies used quotes to support their analysis. However, for 8 papers it was unclear if an adequate range of participants’ views were represented. This was due to the lack of percentages for how many participants the themes related to. The main weaknesses identified were the lack of a clear statement of the researchers’ cultural or theoretical background (n=8), and a lack of addressing the potential influence of the researcher on the research (n=8). The full strengths and weaknesses of individual papers according to the JBI Checklist for Qualitative Research are presented in appendix 1.8.

Discussion

The aims of this review were to synthesise and quality appraise studies of patients' experiences of transition to adult CHD care.

The review highlighted that patients wanted to be actively involved in their transition but at a pace that was individualised to their wants and developmental needs. They wanted to be considered as a whole person, with interest shown beyond their heart condition. This in turn was described to facilitate personal connections and trust with HCPs. Overall, the findings highlighted the importance of patient-centred care within personal and professional relationships and processes before, during and after transition.

This review found strengths and weaknesses in healthcare that facilitated or hindered positive experiences. The general narrative of participants' experiences within paediatric care was more of a personal connection with HCPs who were more likely to show interest in the whole person. This was regarded as lacking in initial contact with adult services, a transition point at which some participants articulated a desired increase in active involvement in their own care. The transition to adult services was more likely to be perceived as positive when the individual felt heard and was given choice in healthcare and transfer decisions. An area of improvement for transition particularly highlighted by patients was more tailored opportunities for education. The review identified the importance of relevant, accessible and timely knowledge in terms of the patient's confidence, level of involvement and responsibility, and successful adjustment to adult services. Specifically, participants emphasised the need for more information about the long-term impact and prognosis of their condition.

Many aspects of patient-centred care that were described can be placed within a developmental framework. Transition occurs during adolescence, a time of development that includes individuation and separation towards young adulthood, where adolescents seek to increase autonomy, place increased

importance on peer connections, and show increased risk-taking. Such stages can be seen in the review's findings including a desire for more information around alcohol and drug use and perceived helpfulness of peer shared experiences. Although dependence on parents decreases in adolescence, parental support constitutes a safe base at times of uncertainty. This is reflected in some participants' appreciation for continued parental support.

Some themes identified in this review echo those identified by Heery and colleagues (2015) including insufficient opportunities for individualised knowledge to facilitate adequate transition, the desire for continuity of person-centred care and for the inclusion of parents within a supportive role. The current review adds additional findings due to the inclusion of research studies that explored young adults' experiences post-transfer (Asp, Bratt, & Bramhagen, 2015; Catena et al., 2018; Coyne et al., 2019; Keir et al., 2018; Lopez et al., 2015). These studies highlighted that similar themes continue into this phase of transition and influence the individual's confidence and ability to adjust. There was also a recognition that insufficient opportunities for education during adolescence became a barrier to participation in adult healthcare decisions. Finally, the need for opportunities to discuss the impact of living with a chronic illness on mental health was highlighted.

Although the focus of this review was CHD, many components of the transition process are similar to a range of paediatric-onset chronic health conditions and require consistent clinical factors for best practice including assessments of 'readiness', support during gradual transition, and the involvement of parents (Aldiss et al., 2015). These factors for best practice are highlighted in the findings of this review. Systematic reviews focusing on the healthcare transition needs and experiences of adolescents and young people with health conditions such as type 1 diabetes, juvenile arthritis and cystic fibrosis have highlighted similar individual variation, thus emphasising the need for tailored, patient-centred care (Coyne et al., 2017; Sheehan et al., 2015; Prior et al., 2014). In

addition, adolescents had increased anxiety when there was a lack of involvement in preparation and transition care, with a lack of opportunity leading to difficulties developing self-management and advocacy skills needed to engage with adult services (Sheehan et al, 2015). Specific to CHD, it is important to acknowledge that a key disease-specific characteristic for many adolescents is that they do not require daily medication, unlike others such as diabetes and cystic fibrosis, and may be asymptomatic. As a result, they may underestimate the severity of their condition and place less importance on continued review and monitoring (Mackie et al., 2019). This emphasises the need for adequate education opportunities during transition and prior to transfer to increase the likelihood of engagement with adult CHD services.

Implications for transition programmes

The review highlights that at the heart of healthcare should be the patient, their views and wishes, and their developmental age and stage. As significant heterogeneity exists, transition should be personalised in accordance with chronological age as well as developmental stage and abilities. To achieve this, care providers' training should incorporate knowledge of developmental stages and skills in adapting clinical practice accordingly. Recognition that the transfer to adult care is a continued period of adjustment, adult services should continue to implement tailored, developmentally appropriate care. For example, adult services should provide scope for parental involvement in line with the young adults' preferences. HCPs need to remain mindful that adolescents are experiencing transition in a broad context and should acknowledge the whole person and multiple transitions that will be occurring.

This review indicates some patients perceive they have insufficient knowledge of their heart condition, which impacts on their ability to actively participate in their healthcare. As such, structured transition programmes may be beneficial. A structured approach facilitates evaluation of the individual as they progress, to

ensure key areas of knowledge and skills are covered. To be tailored to the adolescent's developmental and cognitive stage, education could be provided in multiple formats including verbal, written and online content, as well as formal patient and family education events. For example, in Canada transition programmes have incorporated a 'MyHealth Passport', a portable health summary for patients. Used in combination with a short structured teaching programme, it has been shown to significantly increase and sustain CHD knowledge at 6- and 18-months post-intervention (Mackie et al., 2014, 2018). As adolescence is a time where peer connections are important, involving transition experienced peers may be advantageous. As each CHD patient has a unique medical and developmental history, implementation of standardised transition programmes may be challenging. However, a flexible approach to when and how each patient begins each phase would help to overcome some challenges.

In a Scottish context, it is positive that the standards for transition (Scottish Congenital Cardiac Service, 2018) incorporate aspects of themes highlighted in this review. This includes providing tailored, age-appropriate care within a pathway which incorporates the views of the patient as well as offering peer support. In addition, the standards state that there should be no fixed age of transition but it should be initiated by the age of 14. These standards are intended to be reached by the end of 2020. This review emphasises the importance of meeting this target. Limited resources and an increasing CHD population may hinder the implementation of structured transition programmes in line with this review and the published standards. However, by implementing adequate transition programmes and facilitating transfer, patients are more likely to engage with services in a timely manner, reducing lapses in care and reducing associated negative outcomes.

Quality Appraisal of Existing Research

The quality appraisal identified a similar pattern of strengths and weaknesses for a large majority of the studies. A specific strength of all papers was the inclusion of quotes to aid the illustration of data and clear congruency to conclusions. This in turn increased the transparency of findings and interpretations. The most common weakness was an underreporting of the theoretical and cultural stance of researchers and their potential influence on the research. This has the opposite effect in reducing transparency and authenticity.

It is widely acknowledged that methodological and procedural details are typically underreported in qualitative research, at times due to publication restrictions, and their absence from a manuscript may not reflect their absence from the conduct of the research (Majid & Vanstone, 2018). However, it has been highlighted that researcher reflexivity is a key tenet of qualitative research (Walsh & Downe, 2006). This underscores the importance of reflexive content, including the role of prior assumptions and experience, enhancing the credibility of the research and conclusions. In the context of this review, as underreporting of reflexivity was identified in the majority of papers, results should be interpreted with a degree of caution.

The JBI was selected based on its alignments with the aims and assumptions of this synthesis including its emphasis on researcher reflexivity, application to all qualitative methodologies, and its development from evidence base (Majid & Vanstone, 2018). However, the JBI has less focus on assessing the integrity, transferability and transparency of findings. As discussed by Hannes and colleagues (2010), the JBI tool also does not include a criterion that facilitates the assessment of external validity of original studies in order to provide insight into the transferability of findings to readers. Due to the lack of consensus around assessing quality criteria, a strengths and weakness approach was taken within this review and no papers were excluded from synthesis. Caution therefore needs to be taken when interpreting the results due to the potential

for findings from lower quality studies to be included in analysis. Findings however have suggested that ‘inadequate’ or lower quality studies are less likely to inherently contribute to unique themes or offer any original perspectives on any identified theme (Carroll et al., 2012; Franzel et al., 2013). Overall these limitations highlight the current challenges when using appraisal tools to assess the complex matter of quality and rigor in qualitative research.

Limitations

The search strategy only included English language articles for resource reasons, therefore potentially missing some relevant data and limiting the transferability of the findings to other cultures. The review hoped to incorporate qualitative research on experiences of structured transition programmes to enhance previous systematic reviews. The one article included where some participants undertook a structured transition programme (Catena et al., 2018) did not report these findings separately in order to preserve anonymity, thus eliminating the opportunities to compare qualitative experiences with participants who did not undertake such programmes. In addition, another article that was identified did not meet inclusion criteria as it was not published in English (Thomet et al., 2018). Through grey literature searches and communication with prominent authors, the researcher was made aware of the ongoing qualitative data collection with participants within the STEPSTONES Project (Saarijärvi et al., 2019), a randomised controlled trial with the aim to evaluate the effectiveness of a structured person-centred CHD transition programme. Inclusion of these studies in future reviews would expand the current evidence base.

A potential limitation of the review is also the inclusion of Keir and colleagues’ (2018) research. Although it met the inclusion criteria, the study’s sample was considerably different to other included papers, with an age range of 38 to 49 years old, where participants would have undergone the transition from paediatric to adult CHD care approximately 20 to 30 years prior to participating

in the study. Memory of the experience may therefore be affected due to the length of time passed but may also be more likely to include biases such as selective memory, telescoping or attribution biases. In addition, participants would have experienced transition prior to the publication of best practice guidelines in 2001 (Forbes et al., 2001; Foster et al., 2001) and therefore may have experienced a qualitatively different transition process compared to the collective sample of other included papers. Finally, limitations that have been previously discussed regarding the quality assessment of included research and the use of the JBI should be acknowledged.

Methodological Rigour

The synthesis of qualitative findings is subject to researchers' interpretations including prior biases and assumptions. Use of a reflective log, research supervision, co-ratings of assessing quality, and transparency in reporting enhanced rigour.

Conclusion

This review highlighted the importance of patient-centred care during transition and transfer by consolidating important principles of best practice for adolescents and young adults. Of particular importance was the need for active involvement in their transition at a pace that was individualised to their developmental needs. The need for a holistic approach was noted to facilitate personal connections and trust with HCPs. Best on these principles of best practice, the review suggests implications for transition programmes including a need for flexibility alongside training that incorporates knowledge of developmental stages and skills in adapting clinical practice accordingly.

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Chapter Two: Major Research Project

An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life.

Evelyn Marscheider
BSc (Hons), MSc

Submitted in partial fulfilment of the requirements for the degree of Doctorate
in Clinical Psychology

Address for correspondence:

Evelyn Marscheider / Dr Naomi White

Institute of Health and Wellbeing

College of Medical, Veterinary and Life Sciences

University of Glasgow

Administration Building, Gartnavel Royal Hospital

1055 Great Western Road, Glasgow, G12 0XH

Email: e.marscheider.1@research.gla.ac.uk, naomi.white@glasgow.ac.uk

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 15310 (including appendices)

Plain English Summary

Exploring the experiences of mothers whose infants have congenital heart disease and have undergone surgery in the first year of life.

Background

Congenital Heart Disease (CHD) is a general term for a range of birth defects of the heart. Infants often need specialist care with surgery soon after birth. Research suggests that mothers of children with CHD experience increased levels of distress (Jackson et al., 2015), which is higher if surgery is required during their first year of life (Franck et al., 2010). With a better understanding of mothers' experiences during this time, services may be improved to help manage increased levels of distress.

Aim

This study aimed to explore the experiences of mothers whose infants had a diagnosis of CHD and had surgery within their first year of life.

Methods

5 mothers were recruited from Glasgow's Royal Hospital for Children. Paediatric clinicians helped to identify which mothers were suitable to contact. The researcher contacted mothers who were interested and arranged interviews. The interviews were recorded and analysed to explore themes that the mothers reported.

Results

One main theme was developed to portray the experiences of the mothers in this study: 'A battle on multiple fronts'. 5 subthemes described key parts of the battle: The Trauma of the Battle - "Fighting an uphill battle"; The Fight for Survival - "Is it compatible with life?"; A Fight for Family-Centred Care - "A constant battle to be her parent"; Allies - "Everyone is on the same side"; and A Lonely Battle - "They are not there to help me".

Conclusion

The findings demonstrate that mothers experience an expected battle as a result of receiving an antenatal diagnosis of CHD for their infant. Throughout this fight mothers however described a range of factors that either increased or calmed their sense of threat and conflict. Such factors included support from healthcare professionals, uncertainty and being heard and involved in their infants' care. The findings of this study highlight the importance of healthcare creating a sense of support and togetherness rather than being against that creates more stress for the mothers.

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Plain English Summary Word Count: 398 (including references)

Abstract

Background

The term “congenital heart disease’ (CHD) refers to abnormalities of the heart that are present from birth, and typically require timely medical intervention. The unique stressors faced by mothers whose children have CHD can cause increased psychological distress, impact on quality of life and the capacity for optimal parenting. More research is needed into the lived experiences of mothers whose infants are diagnosed with CHD and undergo surgery to help understand their experiences and target interventions appropriately.

Aim

To explore the lived experiences of mothers whose infants received an antenatal diagnosis of CHD and who had undergone surgery within the first year of life.

Method

Individual interviews were conducted with 5 mothers to explore their lived experiences. All interviews were transcribed verbatim and were analysed using Interpretative Phenomenological Analysis.

Results

One superordinate theme was developed to capture the mothers’ experiences overall: ‘A battle on multiple fronts’. 5 subthemes illustrated the mothers’ battles: The Trauma of the Battle - “Fighting an uphill battle”; The Fight for Survival - “Is it compatible with life?”; A Fight for Family-Centred Care - “A constant battle to be her parent”; Allies - “Everyone is on the same side”; and A Lonely Battle - “They are not there to help me”.

Conclusion

Mothers described an inherent fight following an antenatal diagnosis of CHD, throughout which factors exacerbated or mitigated their sense of threat and conflict. Healthcare services need to create a sense of support and togetherness to reduce the experience of additional stressors in the mothers’ battle.

Background

Congenital Heart Disease

The term “congenital heart disease’ (CHD) refers to abnormalities of the heart that are present from birth and typically require medical intervention soon after birth and follow-up care for life. Cardiac conditions comprise the greatest proportion of all birth defects, are a major cause of foetal and infant mortality, and are the most common group of structural malformations in children (Lissauer, Clayden, & Craft, 2012). Recent advances in treatment has led to the transformation of CHD from a terminal to a chronic condition. With an increased CHD population there has been a shift in focus from survivorship to improving psychological outcomes and quality of life for children and their parents.

Experiences of Parents

Compared to healthy children and children with other conditions, CHD can provide parents with unique stressors of which have been associated with greater levels of psychological symptoms including anxiety and depression (Lawoko & Soares, 2002), anger and hopelessness (Jackson et al., 2015). Such symptoms can impact parents throughout the continuum of their child’s health care experience (Soulvie et al., 2012), with incidents of experiencing trauma symptoms consistent with acute stress disorder (Franich-Ray et al., 2013). Parental stress has been found to be significantly higher when their child underwent cardiac surgery during the first year of life than compared to all other ages (Franck et al., 2010). Increased parental stress during their child’s hospitalisation has been associated with poorer psychological outcomes for the child post-discharge (Kazak et al., 2006). In addition, continued difficulties may impact quality of life and capacity for optimal parenting and responsiveness.

Experiences of Mothers

Recent studies have indicated gender differences between the experiences of mothers and fathers (Jackson et al., 2015). Fathers have been found to focus more on practical tasks and containing emotions (Gower et al., 2017),

experiencing the need to protect their wife and child (Wei et al., 2016), and expressing difficulties balancing employment and support (Sood et al., 2018). Mothers have been reported to experience a higher level of stress than fathers at the time of diagnosis (Bevilacqua et al., 2013) and prior to their child's heart surgery (Utens et al., 2000).

Several qualitative studies have developed our understanding of mothers' experiences (Bruce, Lilja, & Sundin, 2014; Harvey et al., 2013; Lan, Mu, & Hsieh, 2007; Sabzevari et al., 2016). These studies included mothers of children with various CHD diagnoses and at different time points in their child's health care experience. Despite varied focuses, common themes emerged from these studies including reports of feeling a range of intense fluctuating emotions, difficulties with uncertainty and the unknown, difficulties navigating the medical world, trying to mother through it all, and the importance of support throughout. Critical time points of a mother's experience include time of diagnosis, handing their child over to the surgical team, during surgery, and visiting their child in the paediatric intensive care unit after surgery (Wei et al., 2016).

Experiences of Mothers at Diagnosis

Due to advances in foetal ultrasound, there has been an increase in the rates of early detection of CHD during the antenatal period. Research findings have been inconsistent with regards to a difference in impact between antenatal and postnatal diagnosis (Kolaitis, Meentken, & Utens, 2017). Some research has indicated that those who received an antenatal diagnosis had a higher quality of life (Fonseca, Nazaré, & Canavarro, 2012) and had lower anxiety and stress than those diagnosed postnatally (Pinto et al., 2016). Conversely, research has also found no differences between the groups (Bevilacqua et al., 2013; Brosig et al., 2007). However, Bevilacqua and colleagues found that mothers who received the diagnosis prenatally were more depressed, while those who received a postnatal diagnosis were more stressed. This may be related to research that found an antenatal diagnosis gave parents more time to prepare for their journey after

the birth (Sharland, 2012) and gave increased time to process and develop their understanding of the diagnosis than compared to receiving a postnatal diagnosis (Reid & Gaskin, 2018).

Aims of the Current Study

This study aimed to explore the lived experiences of mothers whose infants were given an antenatal diagnosis of CHD, and who underwent surgery within the first year of life. There are very few published studies that explored the experiences of mothers during the full course of their infant's antenatal diagnosis of CHD (Sood et al., 2018). In addition, few studies have been completed with a British population (Gaskin, 2018; Gower et al., 2017; Reid & Gaskin, 2018) with the author not being aware of any published studies focusing on specifically mothers who have received an antenatal diagnosis and their experiences from diagnosis, through birth, subsequent surgery, hospital admission, and discharge home. It is hoped that the rich information gathered will enhance our understanding of the experiences of this group of mothers and provide insight into potential service improvements that may help to manage increased psychological distress.

Method

Recruitment

Ethical approval was obtained (appendixes 2.1 and 2.2). In line with IPA's theoretical underpinnings, particularly the commitment to idiographic principles, purposeful sampling was dependent on the richness and depth of individual accounts (Pietkiewicz & Smith, 2012). The study therefore recruited a small sample of mothers to allow in-depth analysis.

Inclusion and Exclusion Criteria

A main inclusion criterion for this study was the presence of an antenatal diagnosis of CHD. Although research findings have been inconsistent, it is likely that mothers receiving an antenatal diagnosis would experience a different trajectory than mothers postnatally. To obtain a homogeneous sample consistent with IPA, only mothers who received an antenatal diagnosis were included. Other inclusion criteria included:

- Mothers with an infant less than 1 year old
- Their infant had undergone at least one surgical procedure due to a major heart defect at Glasgow's Royal Hospital for Children
- The mother was able to communicate their experiences verbally in English

Following initial recruitment contact, the researcher liaised with relevant HCPs prior to further contact with mothers opting into the research, to discuss whether it was appropriate for the mother to be approached in order to arrange an interview. The decision considered factors such as the outcome of the surgery, the child's health, and the child's prognosis.

Recruitment Procedures

Mothers were identified by HCPs within the cardiology department at Glasgow's Royal Hospital for Children. Any mother who met the inclusion criteria was introduced to the research at a point of routine clinical contact. A clinician known to the family provided mothers with a participant information leaflet outlining the purpose and methodology of the study. It also included information

about confidentiality and the voluntary nature of the study (appendix 2.3). Mothers who indicated an interest were asked to complete a response form (appendix 2.4). The mothers were then contacted by the main researcher to discuss the study further.

Ethical considerations

It was recognised that mothers were going through an emotionally vulnerable time. Steps were taken to reduce any undue burden or distress including the structure of the interview, time taken to build a rapport prior to interview, and management of distress in line with the researcher's clinical experience. Mothers were also signposted to support services. Debriefing and clinical supervision provided reflection space for the researcher to discuss any potential impact of the mothers' experiences and expressions of emotions.

Participants

All mothers identified consented to further information about the research. 9 mothers opted into the research with 6 mothers agreeing to arrange interviews, a response rate of 66.7%. It was mutually agreed for one mother that an interview was inappropriate due to the infant's deteriorating health. Five mothers therefore participated. The mothers were aged from 26-38 years (mean: 32, sd: 4), and their infants ranged in age from 6-45 weeks (mean: 31, sd: 13.3). The length of hospitalisation ranged from 3-39 weeks (mean: 16.2, sd: 14.6), with all infants discharged at the time of interviews. Additional demographic and contextual information are detailed in table 1 below. In order to ensure anonymity, only non-identifiable information has been included when summarising the demographic information of the mothers. Potential identifiable information such as child's diagnosis and health board of residency have been reported separately in table 2. In addition, pseudonyms have not been recorded alongside demographic information in order to reduce the possibility for healthcare workers, who worked closely with the families, to identify the mothers from the illustrative quotations.

Table 1 <i>Demographic Information</i>						
<u>Mother</u>	<u>Age</u>	<u>No. of children</u>	<u>Employment status</u>	<u>Relationship status</u>	<u>Infant's age at time of interview</u>	<u>Time since infant's discharge</u>
1	34	2	Employed	Married	36 weeks	52 days
2	38	2	Self-employed	Married	6 weeks	22 days
3	30	2	Employed	Married	45 weeks	38 days
4	32	2	Employed	Married	31 weeks	148 days
5	26	1	Employed	Co-habiting	37 weeks	253 days

Table 2 <i>Demographic information cont.</i>		
		<u>No. of mothers</u>
NHS Health Board of residency	NHS Lanarkshire	2
	NHS Fife	1
	NHS Ayrshire & Arran	1
	NHS Forth Valley	1
Infant's Diagnosis	Tetralogy of Fallot	2
	Transposition of Great Arteries	1
	Aortic Coarctation VSD	2

Procedure

The main researcher conducted interviews in a clinic room within Glasgow Children's Hospital. Written consent was gained at the start of the interviews (appendix 2.4) and a family information form obtained (appendix 2.5). Interviews lasted 45-90 minutes (mean=66.2minutes; sd: 16.7). A semi-structured interview schedule informed by existing literature and consultation with professionals (appendix 2.6) was used flexibly, including prompts. Interviews were audio recorded digitally and transcribed verbatim on an encrypted laptop. To increase

anonymity, mothers were given a pseudonym and any references that might compromise anonymity were removed.

Data Analysis

IPA was adopted to offer insight into mothers' experiences as it allows a detailed and flexible examination of individuals' lived experiences and their attempts to make sense of their experiences. Transcripts were analysed using a six stage IPA process (Smith et al., 2009). Initial immersion in the data was followed by coding of transcripts, including consideration of descriptive, linguistic and conceptual content (see appendix 2.7). The researcher analysed each transcript individually, identifying emergent themes followed by the construction of superordinate themes for each participant. A table of illustrations and the development of a visual representation was completed to aid this process (appendix 2.8). Themes were then constructed across cases (appendix 2.9). Research supervisors read a sample of transcripts and engaged in follow-up discussions regarding initial codes, themes and emotional hot points of the individual transcripts. Three supervisors read transcript 1, two supervisors read transcript 2, and one supervisor read transcript 3. Analysis was also discussed in detail in frequent academic supervision. The study adhered to the consolidated criteria for reporting qualitative research (COREQ) checklist (Tong, Sainsbury, & Craig, 2007); see appendix 2.10. Quotations have been reported to illustrate the findings. Each quotation was allocated the participant pseudonym and transcript line number to aid transparency of analysis.

Researcher Reflexivity

The development of themes required interpretation of the researcher. As a trainee clinical psychologist, the researcher had experience in paediatric psychology but had not worked with mothers with children with heart conditions. On a personal note, reflexivity was focused on potential biases resulting from the researcher's pregnancy during the interviews and analysis. This included biases on qualitative analysis and the mothers' reflections. For

example, the researcher's pregnancy may have impacted on what the mothers chose to disclose. In recognition of these, a reflective journal was kept by the main researcher. This assisted the process of acknowledging and 'bracketing off' beliefs and expectations throughout the process of analysis. Reflections on such biases were also a prominent feature of research supervision to enhance credibility of results. The supervisors of the research had a range of experience with direct expertise within the field, and a separate supervisor with expertise in IPA methodology. As previously stated, research supervisors reviewed a sample of three transcripts and engaged in discussion of developing themes to enhance the credibility of the analysis.

Results

One superordinate theme was developed to encompass mothers' narratives: 'A battle on multiple fronts'. The experience of receiving a diagnosis of CHD for their infant and the proceeding journey was expected to be challenging; however, the mothers described a journey of unexpected 'uphill battles' and 'nightmares' that you "wouldn't wish on your worst enemy" (Emma, 226). Throughout, they used language and examples that highlighted the trauma of the battle, the fight for survival, the fight for family-centred care, and reflections on allies and a sense of isolation. These aspects were elaborated in the form of subthemes and are illustrated in more detail below.

The Trauma of The Battle - "Fighting an uphill battle"

Throughout their reflections, language was used that compared their experience to a traumatic battle. The experience was described as "fighting another battle" (Sophie, 461), "fighting the baby's corner" (Emma, 72), and a "nightmare" (Linda, 350) where the mothers "live in fear" (Sophie, 631), were "on edge all the time" (Claire, 696), and where "nothing can prepare you" (Sophie, 78). The brutality of the battle was implied by use of language such as "torture", "getting battered" (Emma, 176 & 168), "horrendous" (Jen, 82; Sophie, 699; Emma, 48) and "the worst two weeks in my life" (Sophie, 388). Emma stated, "I wouldn't wish it on my worst enemy" (Emma, 226). Jen described how unexpected events could disrupt progress with the force of a 'knock out' blow:

"It's like snakes and ladders [...] because sometimes you think you are going really well and all of a sudden something comes, and it completely knocks you out." Jen, 1104

The imagery used highlighted the unpredictable, uncontrollable nature of the battle where the odds appeared to be stacked against them. In addition, the emotional intensity evokes a sense of the lasting trauma of the fight with mums reflecting months after the experience. For Linda and Sophie, the traumatic

nature of the battle was reflected in psychological symptoms. Linda described flashbacks and intrusive images of her son's operation. She felt as if her "brain was playing with me" (Linda, 237). For Sophie, the symptoms had an impact at work, when triggering memories and a post-traumatic stress response.

"Heard an arrest call and I just froze because it [pause] brought me back to that time when daughter, where all the doctors just ran and I knew it was her. And it was the strangest feeling. I've never experienced it, as if the ground had swallowed me up and I couldn't get out, I couldn't do anything, and all I could hear was a buzzer." Sophie, 659

In addition, Sophie and Emma drew attention to the paediatric hospital environment as a continuation of a battle scene with other extremely sick children.

"It's not really just babies, I mean you are watching big kids, some 18 years, I mean some died [...] They are just pulling curtains, and you know that someone is just like [sigh] some of the sights we seen [...] it's quite a traumatic place to be watching your baby sleep [...] you were just seeing horrendous sights, families like in bit, it was just terrible." Emma, 291

The Fight for Survival - "Is it compatible with life?"

The first fight this group of mothers reflected on was the day of and process of receiving a diagnosis. All mothers were at first given the news that there may be a problem with the infant's heart at their 20-week antenatal scan. This brought uncertainty and instant fear for what they and their infants faced during the pregnancy and beyond. Claire described that she became 'instantly scared' to talk of the future. As the only first-time mother in this research, Claire also highlighted her sense of injustice around losing the excitement of becoming a mother. All mothers described thoughts that arose about the 'compatibility with life' and worries of losing the infant.

“I was scared to buy things for baby so we kept his room empty [...] I felt that I was pregnant but at the same time I felt so worried that, that we will lose him.” Linda, 102

The delay in a clear diagnosis, due to requiring specialist scans in Glasgow, caused increased uncertainty and distress as the mothers found it hard to not know what was ‘facing’ them and felt they were being ‘battered’ by potential problems. Emma reflected on the psychological impact of her desperation for doctors to provide some reassurance for the life of her unborn infant. Her reflections demonstrate the struggles these mothers faced with having hope during this phase of diagnosis.

“That was just horrendous because you are just waiting for them to say [pause] all I wanted them to say was that they would try and help him but if all these things came back obviously they wouldn’t because they couldn’t [...] it was just [pause] just terrible.” Emma, 47

The nature of the condition itself resulted in the need for surgery. The initial thought of surgery was a challenge for all of the mothers. Sophie’s reflection illustrates the perceived vulnerability of the infants in their fighting to survive an unfair battle.

“It was just really difficult fact to get your head around that this tiny baby would be put through such a huge operation.” Sophie, 64

All mothers expected this part of the fight to be difficult however they felt that nothing could prepare them for seeing their infant for the first time after surgery.

“One nurse in particular who had been a PICU nurse, so she had told us he’ll be in bits, it’ll be the worst. Her words were this will be the sickest that you will ever see them. So we did know that but [sigh] it’s just the worst thing [...] He was [sigh] a pin cushion basically.” Emma, 274

A Fight for Family-Centred Care - “A constant battle to be her parent”

The mothers spoke of a sense of fighting for their infant during their experience. This related not only to the infant’s inability to fight for themselves in terms of being unable to voice concerns with their condition or care, but also in relation to the unique role of a mother to protect and advocate for the best care for their infant. Their reflections also highlighted a sense of fighting to be heard within a medical environment and the conflicts that arose between them and HCPs due to a lack of being heard or involved.

“It felt like a constant battle to be her parent in [local hospital]. They very much made me feel like I was her visitor. Like they completely [pause] disengaged me from her care as much as they could. Which I found really, really difficult.” Jen, 576

“I think mum understand baby better even than them [nurses]. They see only the numbers.” Linda, 460

“The staff didn’t listen to us. Erm, they made decisions without informing me [...] I want to be involved in all her care really you know. I’m her voice. I know her better than anyone.” Sophie, 394

“Being a non-medical person, trying to give medical people advice on how you think is really hard, when sometimes you are right.” Claire, 673

A moment of the battle where fighting for family-centred care appeared to become harder for most mothers was the transition from intensive care to the cardiac ward where the model of care moved away from 1:1 nursing. The change in care appeared to trigger a protective response for the mothers with increased anxiety to leave their infant. For Linda and Claire this resulted in them feeling as if they could not leave their infant alone in the ward and were ‘stuck’ at the infant’s bedside.

“In the PICU there was one nurse with the one baby and over there I remember we came there on the morning, there was one nurse for the one room. We found that very difficult. We were used to, we realised that we won’t be able to leave him alone.” Linda, 351

“Although I never wanted to leave, sometimes you need to leave and we couldn’t. Like I could not leave him in that room, in that bed on his own [...] so we never left.” Claire, 532

Allies - “Everyone is on the same side”

When each mother was asked about the strategies and supports they found useful, they often spoke of not knowing what helped them through. However, an important ally that 4 mothers described was their family. This was particularly important for Claire and Emma whose reflections suggested a comradeship throughout their experience. They felt their families were as ‘invested’ in their infants, stating that “right from the scan it was all of us, all in it together.” (Emma, 334). However, Emma also reflected on the perceived fight she experienced due to her husband struggling to cope with the diagnosis and not being ‘onboard’ with the collective plan. Her husband’s struggle and the impact on her family became an additional fight over and above receiving the diagnosis and her worry for her baby. The difficulty of the experience can be seen in the intensity of emotion expressed.

“At that point [pause] you are trying to fight for, oh I’m going to get upset here [tears], try to fight for your family life that you’ve got [pause] you know, it’s just hard, it’s just hard [...] It got to a point where I was like you either need to get onboard with this plan or you go away, because you are draining me and I don’t have the energy to worry about you and the baby.” Emma, 342

The support of HCPs, especially nurses, was expressed as a vital ally. Emma highlighted the importance of remembering that medical staff were on ‘the same side’ as the parents; although acknowledging this was harder in the heat of the experience.

“Everyone wants what is best for your baby, but at the time you can get a bit, arguing with people because it is not happening quick enough. But you’ve got to remember that everyone is on the same side. Nobody wants anything bad to happen.” Emma, 623

All mothers shared their positive experiences with the nurses in NICU. Claire described that compared to nurses from other departments, the NICU nurses ‘outshone’. She associated their support with not only coping with that stage of the journey but also that their support ‘set her up’ for coping with the entire experience. Jen also spoke of how their support helped her to cope through a difficult time as a patient in the post-natal, labour ward.

“I think if that wasn’t there [support in NICU], I think the whole post-natal bit would have been really, really bad.” Jen, 412

When reflecting on the qualities they felt the NICU nurses had that facilitated a sense of a togetherness, all mothers described the neonatal period as a time where they were encouraged to be involved in their infant’s care, felt heard, and were treated ‘as friends’ with their infants being treated ‘as one of the nurses own’. These descriptions portrayed a sense of belonging where nothing was too much to ask and support was readily available.

A Lonely Battle - “They are not there to help me”

Despite mothers reflecting on the presence and importance of allies such as their family and clinicians, they spoke about a sense of isolation and loneliness

they felt. In relation to their family they described a lack of direct shared experience.

“I’ve got really good support in that sense, you know, if you had to cry you could phone and talk to someone so it was good in that sense. But at the same time, it was almost kind of a barrier, you know that you felt that my baby is going to go through all of this and you don’t understand what we have been told. You know, it’s just really difficult to explain. It was our thing.” Sophie, 90

“It is hard but like [pause] I feel like because me and [partner] know what we have actually been through. Although your family go through it with you like, but being the parents is a different ball game.” Claire, 654

Linda reflected on the impact of culture as a barrier for relying on her family. She spoke of worries, blame and judgements. As a result, she decided to only share the news within her household. For Linda this highlighted the importance of access to additional emotional support from psychological services.

“When I told my mum obviously she knew about the IVF so her first question was is this because of the IVF? And so that kind of question shouldn’t come through, that’s why we don’t want to share with anyone. Rather speak with the psychologist. Would be easier because they won’t be sort of judging you.” Linda, 577

The mothers’ sense of isolation was compounded by perceiving a lack of sought for support; that they were expected to ‘do it themselves’. This was a common theme during the mothers’ time in the labour ward. All mothers highlighted a sense of isolation, of not belonging and therefore not worthy to access care and support. While some experienced anger and frustration as a response, others were accepting that there was no specialist care for them and that as mothers and patients they were an ‘afterthought’.

“You were getting no help whatsoever [...] there was no urgency to help me to get down to [the baby] you know. I actually felt a wee bit inconvenient like we were an inconvenience in there. But I can understand why. They are there to help the mum’s with babies, they are not there to help me.” Emma, 488

“Some of the midwives were really great in helping and others were like well you just need to do it yourself” Jen, 344

In addition, the physical environment of the labour ward added to the sense of isolation. All mothers described being in the same room as other mothers with their babies and claimed that it was one of the hardest parts of their experience.

“That was probably the worst about it, I was in a room with another girl who had her baby [...] she just moaned at her baby all the time [...] like I was sitting there without my baby and I was like [sigh] shut up. Like seriously, I was getting really, that was what was frustrating, it was like I would do anything to have my baby here.” Claire, 299

Discussion

This study utilised IPA to explore the experiences of five mothers with an infant who received an antenatal diagnosis of CHD. One superordinate theme was developed to encompass mothers' narratives: 'A battle on multiple fronts'. 5 subthemes were developed to illustrate the mothers' experiences in more depth in relation to the trauma of the battle, the fight for survival and family-centred, and the experience of allies within a lonely battle.

The mothers' narratives described a journey from expectant joy into an unexpected battle against the heart condition. Although the proceeding journey was expected to be challenging, the mothers described feeling unable to prepare due to high levels of uncertainty and uncontrollability. The emotional intensity mothers expressed throughout the interviews evoked a sense of the lasting trauma. Mothers described a range of psychological symptoms during the experience including being on-edge and flashbacks, consistent with a post-traumatic stress response. The mothers' experiences within the study are consistent with previous research completed by Franich-Ray and colleagues (2013) who found that one-third of mothers experienced trauma symptoms consistent with a diagnosis of acute stress disorder including symptoms of arousal, dissociation and re-experiencing.

As mothers were unable to 'flee' from this battle, they described their 'fight' response to threats that presented. These were heightened by the mother's unique role and instinct to protect their infant by advocating for the best possible, family-centred care. The mother's unique role appeared to be associated with a feeling of isolation, despite support from family or clinicians, due to a lack of shared experience with mothers. The surrounding services were positioned either to represent allies providing support to mitigate the mother's sense of threat and distress, or were positioned to compound their difficulties, representing additional, unnecessary fights that increased distress and isolation. An example of a mitigating ally included HCPs being perceived as 'on the same

side' as the mothers. NICU nurses in particular appeared to facilitate a sense of togetherness and empowerment. In contrast, not feeling heard or involved in their infants' care, feeling as an 'afterthought' without specific care for themselves, and environmental factors such as the labour ward were found to compound the struggle. Similar themes have been found in previous research including challenges with navigating the medical world and dealing with the unknown (Harvey et al., 2013). In addition, Bruce and colleagues research (2014) highlighted the importance of support that was adjusted to the mothers' needs and desires.

All mothers in this research received an antenatal diagnosis. Their experiences highlighted increased levels of uncertainty and anxiety during the diagnosis phase while waiting on test results. Living with uncertainty without reassurance from medical staff for this period of time stood out as particularly challenging and isolating. Although previous research findings have been inconsistent with regards to a difference in impact between antenatal and postnatal diagnosis (Kolaitis et al., 2017), this finding may highlight a difference for some mothers who receive a diagnosis postnatally as some will have the additional support from neonatal staff due to the admission of their infant after birth.

Implications for Services

Services either represented an ally during the mothers' experiences and facilitated coping or compounded the mothers' struggles. The study highlights the importance of communicating a message of solidarity with the mothers, emphasising how they are all on the same side to help the infant fight for survival against the condition, but also to communicate they are also there to provide direct support for the mother, respecting her unique role in the infant's care. It may be helpful for services to consider the implementation of the framework for trauma-informed care (NHS Education for Scotland, 2017), particularly as the mothers are likely to be experiencing ongoing trauma from

the journey of diagnosis to discharge. The framework focuses on implementing key principles including choice, collaboration, trust, empowerment and safety; all of which echoed facilitators for coping described by the mothers in this study. Practical changes may include ensuring that mothers are given opportunities for appropriate involvement in their infant's care, increased preparation for transition to ward based care, and better links and communication between the labour ward and NICU. Small changes to the environment may make a substantial difference in mitigating levels of distress and isolation such as, when possible, reducing the need to share a room with mothers who are caring for their babies in the labour ward.

Limitations

Findings are based on the reflections of five mothers who agreed to be interviewed. The study therefore represents the experiences of this group of mothers and caution should be taken in respect to the transferability. The study has attempted to provide a detailed description of recruitment, data collection, analysis and the study context in order to enable others to judge the transferability of the findings. Although efforts were made to recruit a homogenous sample, the mothers differed in aspects such as CHD diagnosis, length of time before first surgery, and whether they were a first-time mother or not which are likely to have influenced their experiences.

IPA involves adopting a double hermeneutic stance in which the development of themes requires the researcher to play an active role in the interpretation of the participants' own expressed interpretations. In choosing to highlight and elaborate upon certain recurrent themes, it is acknowledged that some nuances may inherently be lost through this process in the pursuit of synthesising the experiences across the whole sample of mothers. As such, the metaphor of a "battle", chosen to convey an important overarching theme, has limitations in

capturing nuances in the complexity of these participants' experiences as a whole.

Future Research

The study highlighted several differences that would be beneficial to research in more depth. This includes the impact of culture on the perceived and accessible support for mothers and the ways healthcare could tailor their approaches to mitigate any increased distress. As previously stated, the approaches and systems within NICU appeared to facilitate a sense of togetherness and empowerment for this group of mothers. A more in-depth understanding of this would be helpful to inform service improvements in other wards and departments. In addition, further research into nurses' experiences of supporting mothers and families would increase the understanding of this dynamic. Finally, this study eluded to the benefits of shared experience for this group of mothers. Research into the potential options and benefits for methods of shared experience such as mentoring, support groups and use of social media or technology may therefore be helpful.

Conclusion

The findings demonstrate the inherent fight that mothers experience following an antenatal diagnosis of CHD for their infant. Throughout this fight, mothers describe a range of factors that can either exacerbate or mitigate the sense of threat and conflict. The findings underscore the value of systems creating a sense of support and togetherness rather inadvertently creating additional stressors in the mother's battle.

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Appendices: Systematic Review

Appendix 1.1: Manuscript Of Submission Guidelines For Journal Of Cardiology In The Young

CAMBRIDGE | Instructions for Contributors

CARDIOLOGY IN THE YOUNG

Submission to *Cardiology in the Young* is exclusively via the web-based peer-review system, [CTY Manuscript Central](#).

Online submission enables rapid review and allows online manuscript tracking. Please use the following URL: <http://mc.manuscriptcentral.com/cty>

Clinical Trials

As a condition of consideration for publication, registration of clinical trials in a public trials registry is required. A clinical trial is defined by the [International Committee of Medical Journal Editors](#) (in accordance with the definition of the World Health Organisation) as any research project that prospectively assigns human participants or groups of humans to one or more health-related interventions to evaluate the effects on health outcomes. Trials must be registered before the start of patient enrolment. The registry must be accessible to the public at no charge. It must be open to all prospective registrants and managed by a not-for-profit organization. There must be a mechanism to ensure the validity of the registration data, and the registry should be electronically searchable. An acceptable registry must include at minimum a unique trial number, trial registration date, secondary identification information if assigned by sponsors or others, funding source(s), primary and secondary sponsor(s), responsible contact person, research contact person, official scientific title of the study, research ethics review, the medical condition being studied, intervention(s), key inclusion and exclusion criteria, study type, anticipated trial start date, target sample size, recruitment status, primary outcome, and key secondary outcomes. Registration information must be provided at the time of submission. **Trial registry name, registration identification number, and the URL for the registry should be included at the end of the abstract.**

Manuscripts reporting the results of randomized controlled trials should include a ["CONSORT"](#) flow diagram to illustrate the progress of all patients in the study (See: Schulz KF, Altman D, for the CONSORT Group. The CONSORT statement: revised recommendations for improving the quality of reports of parallel-group randomized trials. *JAMA*. 2001;285(15):1987–1991.) The flow diagram should be uploaded as a separate file to the manuscript.

Editorial Policies

Cardiology in the Young is devoted to cardiovascular issues affecting the young and the older patient with the sequels of cardiac disease acquired in childhood. Submission of both basic research and clinical papers is encouraged. Articles on fundamental principles will also be considered for publication. Reviews on recent developments are welcome. The Journal serves the interest of all professionals concerned with these topics. By design, the Journal is international and multidisciplinary in its approach, and the members of the Editorial Board take an active role in the Journal's mission. Prospective authors are encouraged to consult with the editors and members of the Editorial Board with any enquiries. The editors encourage the submission of articles from developing countries.

Articles should be concerned with original research not published previously and not being considered for publication elsewhere. Submission of a manuscript to the Journal gives the publisher the right to publish that paper if it is accepted, and the copyright of the manuscript becomes property of the publisher. Manuscripts may be edited to improve clarity and expression.

Authors must ensure that their studies comply with appropriate institutional and national guidelines for ethical matters. Specifically, by submission of a manuscript, the authors are responsible for compliance with guidelines and regulations of the authors' institution and all appropriate governmental agencies.

Articles including human subjects must include a statement that informed consent was obtained and that the study was reviewed and approved by the institution's committee on human experimentation. Articles including animal experimentation must conform to the principles of the American Physiological Society, and a statement acknowledging conformation to these standards must be included in the *Materials and methods* section of the manuscript. Authors are also requested to identify possible conflicts of interest, especially if they relate to commercial sponsorship or equity holdings.

Use of Abbreviations

Abbreviations are **not** allowed in the text except for the following **only**: NYHA, CHD, MRI, CT, ICU and units of measure such as mmHg or kg, are allowed. However, any abbreviations can be used on tables and figures.

Language

The language of the Journal is English, but acceptance of a manuscript will reflect scientific rather than grammatical content. The editors undertake to facilitate the publication of papers from those authors whose native language is not English.

Cambridge recommends that authors have their manuscripts checked by an English language native speaker before submission; this will ensure that submissions are judged at peer review exclusively on academic merit. We list a number of third-party services specialising in language editing and / or translation, and suggest that authors contact as appropriate. Use of any of these services is voluntary, and at the author's own expense.

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Manuscripts

Manuscripts should be submitted via the web-based peer-review system, [CTY Manuscript Central](#) and must include a complete set of Figures. Further information regarding Figure formats is outlined below.

Authorship

Authorship should be assumed only by those workers who have contributed materially to the work and its report, and who accept the responsibility for the accuracy of the concepts expressed. Colleagues who have otherwise assisted or collaborated should be recognized in the section for acknowledgements. An excellent guide to authorship is given by the Style Manual Committee of the Council of Biology Editors, and the editors encourage consultation with this source.

Style

The Journal uses the English language and as such does not use Latin terms such as 'superior vena cava'. Anatomic terms should be given in the English language. Headlines and subheadings should be liberally employed in the methods, results, and discussion sections. Use short paragraphs whenever possible. The authors should strive for clarity of expression, avoiding, in particular, the use of jargon. Authors should also avoid conventions such as Group 1, Group 2, and so on, using descriptive titles rather than alphanumeric codes. Authors should use the definite/indefinite article where required.

Title page

The manuscript should include a title page, and it should follow these rules:

1. Author qualifications (for example MD) must **not** be included on the authors list on the title page, except in the address for correspondence.
2. Include the first names of the authors, followed by the initials of any middle names, and finally, the family name.
3. Include the affiliation address(es) for the authors, to include the department, institute, city and country.
4. Include an address for correspondence, with the full postal address, telephone and e-mail address of the author in question.

Manuscript types

Allowed manuscript types are Original Article, Article Commentaries, Brief Report, Review Article, Editorial, Image, Letter and Guidelines. Each is described below in detail.

Original Articles

Divide the manuscript into the following sections:

- o Title Page (see instructions on page 2)
- o Abstract
- o Keywords (3-6).
- o Headings: Introduction, Materials and methods, Results, Discussion, Acknowledgements, Financial Support, Conflicts of Interest, Ethical Standards, References, Tables, Figure legends and Figures

Abbreviations

Abbreviations are **not** allowed in the text except for the following **only**: NYHA, CHD, MRI, CT, ICU and units of measure such as mmHg or kg, are allowed. However, any abbreviations can be used on tables and figures.

Abstract

The Abstract should be no more than 250 words and should include statements identifying relevant methods and results justifying publication. Do not use abbreviations or symbols.

Introduction

The Introduction should be brief and set out the purposes for which the study has been performed. It should not include an extensive review of the literature.

Materials and Methods

The Materials and Methods should be sufficiently detailed so that readers and reviewers can understand precisely what has been done without studying the references directly. The description may be abbreviated when well-accepted techniques are used with appropriate reference to previously published methods. Statements confirming conformation to institutional and governmental review of the experimental protocol (see above in the Editorial policies section) should be included here.

Results

The results should be presented precisely. Reference to Tables and Figures, to the extent that they contribute substantively to help the reader understand clearly the relevant positive and negative findings, is encouraged. Keep discussion of their importance to a minimum in this section of the manuscript.

Discussion

The Discussion should relate directly to the study being reported. The Discussion should interpret the results, should describe the relevance of the results, and should include a discussion of the limitations of the study. Do not include a general review of the topic.

Acknowledgements

Acknowledgements should follow the discussion. Here you may acknowledge individuals or organisations that provided advice and/or support (non-financial). Formal financial support and funding should be listed in the following section. The Acknowledgements should be placed after the main body of the text before Financial Support. If there are no Acknowledgements, the title should be inserted followed by "None". Papers that do not include an Acknowledgements section will not be reviewed.

Financial Support

The Financial Support statement should be placed after the Acknowledgements and before the Conflicts of Interest section. Please provide details of the sources of financial support for all authors, including grant numbers. This is particularly important in the case of research that is supported by industry. Support from industry not only includes direct financial support for the study but also support in kind such as provision of medications, equipment, kits or reagents without charge or at reduced cost and provision of services such as statistical analysis. For example, "This work was supported by the Medical research Council (grant number XXXXXXXX)". Multiple grant numbers should be separated by a comma and space, and where research was funded by more than one agency the different agencies should be separated by a semi-colon, with "and" before the final funder. Grants held by different authors should be identified as belonging to individual authors by the authors' initials. For example, "This work was supported by the Wellcome Trust (A.B., grant numbers XXXX, YYYY), (C.D., grant number ZZZZ); the Natural Environment Research Council (E.F., grant number FFFF); and the National Institutes of Health (A.B., grant number GGGG), (E.F., grant number HHHH)". Where no specific funding has been provided for research, please provide the following statement: "This research received no specific grant from any funding agency, commercial or not-for-profit sectors." Papers that do not include a Financial Support statement will not be reviewed.

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committees (please name) ." and "The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guides on the care and use of laboratory animals (please name) and has been approved by the institutional committee (please name)." If the research does not involve human and/or animal experimentation, this statement should be omitted. Papers reporting the results of human and/or animal experimentation that do not contain an Ethical Standards statement will not be reviewed. For more information on the ethical standards and procedures of Cambridge Journals, please visit [Cambridge Journals Online](#).

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Case Reports / Brief Reports

Case Reports which add important new information will be published as Brief Reports, and must conform to the following requirements:

- Abbreviations are **not** allowed in the text except for the following **only**: NYHA, CHD, MRI, CT, ICU and units of measure such as mmHg or kg, are allowed. However, any abbreviations can be used on tables and figures.
- Headings before the References: Financial Support, Conflicts of Interest, Ethical Standards (Please see under *Original Article* above for the exact wording).
- A title page (see instructions on page 2)
- A maximum of three authors
- An abstract (of about 60 words)
- A maximum of 1,000 words
- 3-6 key words
- 2 Figures **OR** 2 Tables only
- A maximum of 2 supplementary material files
- A maximum of 10 references.

Editorials and Letters to the Editor

Readers are encouraged to write about any topic that relates to cardiology in the young. Such letters will appear in *Letters to the Editor*. They should be no longer than 500 words. Please include a title page (see instructions on page 2 above).

Editorials are written on invitation but unsolicited articles of approximately 1,500 words which may have particular topical interest will be welcomed for consideration. Please include a title page (see instructions on page 2 above). For Editorials and Letters please also see the note on abbreviations under Images, below.

Images in Congenital Cardiac Disease

Definitive, unique, or extraordinary pictures of any aspect of congenital cardiac disease will be presented and must conform to the following requirements:

- Abbreviations are **not** allowed in the text except for the following **only**: NYHA, CHD, MRI, CT, ICU and units of measure such as mmHg or kg, are allowed. However, any abbreviations can be used on tables and figures.
- Headings before the References: Financial Support, Conflicts of Interest, Ethical Standards (Please see under *Original Article* above for the exact wording).
- A title page (see instructions on page 2)
- A maximum of three authors
- An abstract (of about 30 words)
- A maximum of 250 words
- 3-6 key words
- A maximum of 4 images
- Only 1 reference.

Instructional value and artistic merit will be considered in addition to scientific import and clinical relevance.

Guidelines

CTY welcomes papers that consist of clear, concise guidelines in any technique or methodology that relates to the practice of cardiology and cardiac surgery in the young. Guidelines should be evidence based and should reflect professional consensus of best practice. Generally they will be expected to be endorsed by professional societies and associations. These papers do not follow any specific format, and may include figures and tables. Guidelines may be subject to peer review before acceptance for publication. Please include a title page (see instructions on page 2 above) and please see the note on Abbreviations under Images.

References

References should be numbered consecutively (in superscript) as they appear in the text. Type the reference list with double-spacing on a separate page. References (using Index Medicus abbreviations) should appear in the style as demonstrated below. Please note that if more than six authors, the **first three** authors should be listed and then 'et al.' Examples:

1. Redington AN, Rigby ML, Oldershaw P, Gibson DG, Shinebourne EA. Right ventricular function 10 years after the Mustard operation for transposition of the great arteries: analysis of size, shape, and wall motion. *Br Heart J* 1989; 62: 455-461.
2. Smith VR, Jones AL, Miller W et al. Left ventricular myocardial velocities in children. *Eur Heart J* 2000; 21: 104-112.
3. Zuberbuhler JR. *Clinical Diagnosis in Pediatric Cardiology*. Churchill Livingstone, New York, 1981.
4. Frantz EG. Adult respiratory distress syndrome in children. In: Harried HS, Jr (ed.) *Pediatric Pulmonary Heart Disease*. Little, Brown, Boston, 1990, pp 315-324.

Tables

Tables should follow the style as demonstrated in issues to date, and be essential to the understanding of the text. Tables should have short descriptive titles and should be numbered (1, 2, 3 etc.) as they appear sequentially in the text. If only one Table is included, it should be referred to as Table. Submit each Table on a separate sheet of paper. All abbreviations and symbols should be defined in a footnote below the Table.

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Colour mode: black and white (also known as 1-bit)

Resolution: 1200 dpi

Combination artwork (line/tone)

Format: tif or eps

Colour mode: grayscale (also known as 8-bit)

Resolution: 800 dpi

Black and white halftone artwork

Format: tif

Colour mode: grayscale (also known as 8-bit)

Resolution: 300 dpi

Colour halftone artwork

Format: tif

Colour mode: CMYK colour

Resolution: 300 dpi

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If you do not yet have the copyright, then please say something along the lines of <copyright> pending, etc.

(Revised 14 Dec 2015)

Appendix 1.2: Example Of Full Search Strategy In Medline

Source: Ovid Medline (R) and In-Process & Other Non-Indexed Citations (1946 to September 10, 2019)

Interface: Ovid - access through The University of Glasgow

Search date: 11 September 2019

Search strategy:

1	exp Heart Defects, Congenital/	146483
2	("congenital heart" or "heart defect*" or "heart disease*" or "heart syndrome" or "congenital abnormalities" or "cardiovascular abnormalities").tw.	176602
3	("heart" or "cardiac").tw.	1108441
4	("disorder*" or "abnormalit*" or "defect" or "disease").tw.	4010342
5	3 and 4	354259
6	1 or 2 or 5	479158
7	exp "Continuity of Patient Care"/ or exp Transition to Adult Care/ or exp Patient Transfer/	231368
8	("transition*" or "transition care*" or "patient handoff" or "patient hand-off" or "transfer*" or "retention" or "continuity*" or "move*" or "moving" or "hand-over").tw.	1544057
9	7 or 8	1747886
10	exp Qualitative Research/	48175
11	("qualitative" or "experience*" or "interview" or "explor*" or "perspective*" or "theme*").tw.	2161853
12	10 or 11	2166906
13	6 and 9 and 12	2653
14	Limit 13 to yr="2001-Current"	<u>2148</u>

Appendix 1.4: Process Of Thematic Synthesis

<i>Thomas & Harden (2008) stages</i>	<i>Description</i>
STAGE 1: Free line-by-line coding of the findings of primary studies	<ul style="list-style-type: none"> • Enter the verbatim findings of studies into database • Code each line of text according to its meaning and content • Codes structured as ‘free’ codes without a hierarchical structure • Examine text with the same code to check consistency
STAGE 2: Organisation of ‘free codes’ into related areas to construct ‘descriptive’ themes	<ul style="list-style-type: none"> • Look for similarities and differences between codes to begin grouping into a hierarchical tree structure • Create new codes to capture meaning of groups of ‘free codes’
STAGE 3: Development of ‘analytical’ themes	<ul style="list-style-type: none"> • Use descriptive themes that emerged to explore research aim and questions • Create analytical themes that capture the descriptive themes in the context of questions e.g. considering the implications for intervention development

Appendix 1.5: Example of Analytical Theme

<i>Analytical themes</i>		
<u>Analytical theme</u>	<u>Subthemes</u>	<u>Illustrations</u>
<p>"I was given a choice and I liked that" - opportunities for participation</p>	<p>Involvement / participation</p>	<p>"The most important thing to me when transferring to the adult clinic was that all the decisions were made with me involved and I was able to talk about what was going to happen when I saw the doctors, which I never had a chance to do in the children's clinic." (Wray et al., 2008, p571). "I think it was just the last appointment at outpatients. It was thanks very much, 'bye, bye, best of luck'. It's just the way it was done. There was no 'come in and we'll have a chat about this and what do you think and how would you feel.'" (Coyne et al., 2019, p4067). "He [paediatric cardiologist] suddenly said, this is your last check-up in the paediatric cardiac unit and they hadn't said anything about it before. So it was very sudden." (Asp et al., 2015, p5). "My only gripe is that I didn't know what was going to happen in advance and I did find that a bit awkward [...] it was all a bit unexpected." (Moons et al., 2009, p319). "Being treated like an adult in the adult clinic is wonderful and getting more involved and taken seriously feels good, it's a huge difference." (Asp et al 2015) "I feel that I must become more and more involved after the transfer, as it's about my body and I know that my parents can't be there to obtain the information and thus I have to take it in" (Asp et al 2015)</p>
<p>Opportunities for knowledge</p>	<p>Opportunities for knowledge</p>	<p>"I want to know everything that there is to know about my heart condition [...] cause I was so young when I had surgery to know what was going on, but now</p>
		<p><u>Representations</u></p> <p>Asp Burström Catena Coyne Keir Moola Moons Wray</p>

that I am older, I want to understand what is going on with me, and to understand what might happen in the future with me.” (Moola, 2011, p845)
“I’m getting older. I feel more comfortable voicing my concerns and asking them more questions, like more specific questions and I feel like I have a right to know as opposed to when I was younger, I kind of just listened and agreed with whatever they said but I feel like now I don’t want to say demanding but I, expecting more from my visits.” (Catena et al 2018)

Being heard “It feels as if you are involved and have a say and if you are unhappy about something, they take note of it, check it up and phone back.” (Asp et al 2015)

Choice “I was given a choice and I liked that because it made me feel that I was getting older and was given more responsibility.” (Asp et al., 2015, p6).
“My mother just took control. She’s like that ‘cause I’m the youngest, and the heart condition, I don’t like that.” (Catena et al., 2018, p595).
“She says I’m forgetful, so she wants to come and hear stuff from the doctors herself. But I keep telling her that she had to let me go sometimes just by myself.” (Catena et al., 2018 p595).
“I felt forced the first time they said that I must go alone without my parents. I refused because I wanted to have my parents with me.” (Burstrom et al, 2017)
“Got shoved right into it” felt experience would have been better if he
“Could’ve just stayed back and slowly got into it” (Catena et al, 2018)

Impact It’s hard to take ownership because I didn’t know what my condition was. (Keir et al., 2018, p745).

Appendix 1.6: Breakdown of Themes

<i>Breakdown and presence of themes in articles</i>			
	Being an active participant in individualised care		
	“I was given a choice and I liked that” - <u>opportunities for involvement</u>	“It was like they had to tell me everything, it was horrible!” - <u>individualised opportunities</u>	“Take in the whole person” - <u>the need for holistic care</u>
Asp, 2015	X	X	X
Burström, 2016		X	X
Burström, 2017	X	X	
Catena, 2018	X	X	X
Coyne, 2019	X	X	X
Keir, 2018	X	X	
Lopez, 2015		X	
Moola, 2011	X	X	
Moons, 2009	X	X	X
Wray, 2008	X	X	X

Appendix 1.7: Quality Appraisal Framework - JBI Critical Appraisal Checklist For Qualitative Research

JBI Critical Appraisal Checklist for Qualitative Research

Reviewer _____ Date _____

Author _____ Year _____ Record Number _____

	Yes	No	Unclear	Not applicable
1. Is there congruity between the stated philosophical perspective and the research methodology?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
2. Is there congruity between the research methodology and the research question or objectives?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
3. Is there congruity between the research methodology and the methods used to collect data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
4. Is there congruity between the research methodology and the representation and analysis of data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
5. Is there congruity between the research methodology and the interpretation of results?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
6. Is there a statement locating the researcher culturally or theoretically?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
7. Is the influence of the researcher on the research, and vice-versa, addressed?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
8. Are participants, and their voices, adequately represented?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
9. Is the research ethical according to current criteria or, for recent studies, and is there evidence of ethical approval by an appropriate body?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>
10. Do the conclusions drawn in the research report flow from the analysis, or interpretation, of the data?	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>	<input type="checkbox"/>

Overall appraisal: Include Exclude Seek further info

Comments (Including reason for exclusion)

Appendix 1.8: Quality Appraisal Of Included Studies

<i>Quality Appraisal of Included Studies</i>			
First author, Year	Congruity - philosophy & method	Congruity - method & objective	Congruity - method & data collection
	<u>Strengths</u>	<u>Strengths</u>	<u>Weaknesses</u>
Asp, 2015	Content analysis for descriptive qualitative study with interviews	Content analysis to explore experience	Semi-structure individual face-to-face interviews Open-ended questions at start Participants allowed to freely relate their recollections Structured questions at end but still open-ended Range in duration of interviews (15-53 minutes)
Burström, 2016	Content analysis for exploratory design	Content analysis to identify and describe needs	Semi-structured interviews Interviews considered full and sufficient Large variation in length of interviews
Burström, 2017	Content analysis for qualitative study	Content analysis to explore views	Focus groups No statement indicating flexibility of interview guide
Catena, 2018	Thematic analysis therefore allows for flexible theoretical basis	Thematic analysis to explore perspectives	Semi-structured telephone interviews Unequal sampling for two groups No statement included of flexibility in interviews with set list of questions

Coyne, 2019	Thematic analysis therefore allows for flexible theoretical basis	Thematic analysis to explore experiences of transition	Semi-structured face-face interviews Flexible interview schedule Responsiveness by interviewers to issues raised by participants	30 minute interviews - query richness of data Large sample size - query depth of analysis
Keir, 2018	Narrative analysis established as research method with appropriate analysis with main focus on thematic approach with some structural and dialogue	Narrative analysis to gain insight on sense making of experience	Narrative analysis interview Participants guided interviews with little prompting as necessary	
Lopez, 2015	Appears to be thematic analysis	Appears to be thematic analysis to explore perceived needs and concerns	Semi-structured telephone interviews	Interviews appear more structured with set questions
Moola, 2011	Thematic analysis therefore allows for flexible theoretical basis	Thematic analysis to explore understanding and perspectives	Semi-structured interviews Participants response drove further lines of enquiry 45-90 minutes long	Large sample size - query depth of analysis
Moons, 2009	Phenomenologic approach with in-depth interviews	Phenomenologic approach to explore expectations and experiences	Semi-structured interviews Descriptive question first followed by more in-depth question	Interview duration range not reported
Wray, 2008	Thematic analysis therefore allows for flexible theoretical basis	Thematic analysis to explore views	Postal survey appropriate for preliminary exploratory research	

First author, Year	Congruity - methodology & analysis		Congruity - methodology & interpretation		Locating the researcher culturally or theoretically	
	<u>Strengths</u>	<u>Weaknesses</u>	<u>Strengths</u>	<u>Weaknesses</u>	<u>Strengths</u>	<u>Weaknesses</u>
Asp, 2015	Similarities and differences reported Descriptive accounts		Clear congruency Independent reading of transcripts followed by two levels of consultation Acknowledgement of limits to generalising and also transference to other contexts		Statement of qualification, occupation and experience in relation to CHD	
Burström, 2016	Similarities and differences reported	Limited information on analysis procedures	Clear congruency 3 co-researchers Brief acknowledgement of limitations	Limited discussions on generalisability		Only details of qualifications and place of work for contributors
Burström, 2017	Similarities and differences reported		Clear congruency 3 co-researchers Brief acknowledgement of limitations	Limited discussions on generalisability		Only details of qualifications and place of work for contributors
Catena, 2018	Presentation of analysis congruent with thematic analysis Similarities and differences of the whole sample reported	Analysis not related to main objective of research - to identify similarities and differences of participants in ACHD versus paediatric settings. Focus on general themes	Clear congruency	No acknowledgement of limitations to generalisability		Only details of qualifications and place of work for contributors
Coyne, 2019	Similarities and differences reported Comprehensive analysis pathways		Clear congruency Advisory groups and team meetings to aid interpretation	No discussion of study limitations	Details of experience and training in interviews reported	No further specific statements in experience of qualitative analysis

Keir, 2018	<p>Advisory groups and team meetings to aid analysis</p> <p>Similarities and differences reported</p> <p>Number of participants themes relevant to reported</p> <p>Similarities and differences reported</p> <p>Number of participants themes relevant to reported</p> <p>Similarities and differences reported</p> <p>Detailed field notes and potential analytical insights</p> <p>Clear iterative process</p> <p>Similarities and differences reported</p> <p>Presentation of data consistent with phenomenologic approach</p>	<p>Clear congruency</p> <p>Acknowledgement of limitations to generalisability</p> <p>Congruency of data reported to interpretation</p> <p>Acknowledgement of limitations to study</p>	<p>Set questions appeared to have had some influence on themes</p> <p>Limited information on analysis procedures</p>	<p>Limited information on process of interpretation</p> <p>No discussion of study limitations</p>	<p>Researcher who completed analysis stated to be expert in qualitative analysis</p> <p>Clear paragraph and discussion of theoretical stance</p>	<p>Only details of qualifications and place of work for contributors</p> <p>No statements on theoretical stance throughout</p>
Moola, 2011		<p>Clear congruency</p>		<p>No discussion of study limitations</p>	<p>Clear paragraph and discussion of theoretical stance</p>	
Moons, 2009		<p>Clear congruency</p> <p>Reflective diary kept</p> <p>Discussion and consultation of interpretation</p>			<p>Statement of occupation and experience with CHD</p>	<p>No further specific statements in experience of qualitative analysis</p>
Wray, 2008	<p>Brief overview of themes given consistent with preliminary analysis</p>	<p>Clear congruency</p> <p>Acknowledgement of small study size and impact of context</p>	<p>Focus on similarities</p>	<p>Limited discussion regarding generalizability</p>		<p>Only details of qualifications and place of work for contributors</p>

First author, Year	Influence of researcher		Representation of participants		Ethical approval		Relationship of conclusion to analysis / interpretation	
	<u>Strengths</u>	<u>Weaknesses</u>	<u>Strengths</u>	<u>Weaknesses</u>	<u>Strengths</u>	<u>Weaknesses</u>	<u>Strengths</u>	<u>Weaknesses</u>
Asp, 2015	Acknowledgement of potential influence of researcher's perspectives on interpretation and methodology to limit		Variety of quotes for all themes	No reporting of number of participants themes were relevant to	Advisory statement provided by Regional Ethic Review Board		Clear demonstration of conclusions drawn from data	
Burström, 2016		No reflections or statements included.	Multiple quotes for all themes	No reporting of number of participants themes were relevant to	Statement that ethical approval requested	Unclear if ethical approval was approved	Clear demonstration of conclusions drawn from data	
Burström, 2017		No reflections or statements included.	Multiple quotes and variety of quotes for all themes	No reporting of number of participants themes were relevant to	Statement that conformed to ethical standards		Clear demonstration of conclusions drawn from data	
Catena, 2018		No reflections or statements included. No information given regarding qualifications and experience of individual who	Variety of quotes Use of participants own language	No reporting of number of participants themes were relevant to	Ethical approval obtained for two university-based ethics boards		Clear demonstration of conclusions drawn from data	Conclusions not related to study objective

	completed interviews						
Coyne, 2019	Statement that rapport developed first with participants	No specific statements regarding potential influence of researcher	Good number of quotes for every theme	No reporting of numbers of participants themes were relevant to	Ethical approval obtained for University and Hospital committees	Clear demonstration of conclusions drawn from data	
Keir, 2018	Statement that participants had previously met interviewers in routine medical context	No reflection of potential influence of previous meeting No recognition of researchers on interpretation of narratives	Quotes from range of participants Number of participants themes relevant to reported Use of participants own language	Ethical approval by institutional research ethics board	Clear demonstration of conclusions drawn from data Language of participants quoted and used		
Lopez, 2015	No reflections or statements		Multiple quotes for themes Percentage of participants themes relevant to reported	Ethical approval by Institutional Review Board	Clear demonstration of conclusions drawn from data		
Moola, 2011	Journal of field notes and analytic insights	Unsure if kept in a reflective form No acknowledgement of researchers on interpretation	Multiple quotes given for themes	No reporting of numbers of participants themes were relevant to (particularly important as large sample size)	Ethical approval by Hospital Research Ethics Board	Clear demonstration of conclusions drawn from data	

<p>Moons, 2009</p>	<p>Acknowledgement of potential bias. Reflective diary kept.</p>	<p>Quotes used to illustrate themes</p>	<p>Often only one quote for a theme illustrated No reporting of number of participants themes were relevant to No quotes included for views concerning adult services No reporting of number of participants themes were relevant to</p>	<p>Ethical approval by Institutional Review Board</p>	<p>Clear demonstration of conclusions drawn from data</p>
<p>Wray, 2008</p>	<p>No reflections or statements included.</p>	<p>Quotes used to illustrate for most themes</p>	<p>No quotes included for views concerning adult services No reporting of number of participants themes were relevant to</p>	<p>Ethical approval by Local Research Ethics Committee</p>	<p>Clear demonstration of conclusions drawn from data Conclusions consistent with level and depth of research</p>

Appendices: Major Research Project

Appendix 2.1: Research Ethics Approval

WOSRES
West of Scotland Research Ethics Service



Mrs Evelyn Marscheider
University of Glasgow
1st Floor Administration Building, Gartnavel Royal
Hospital
1055 Great Western Road
G12 0XH

West of Scotland REC 6:
Research Ethics
Ground Floor Ward 11
Dykebar Hospital
Grahamston Road
Falsley PA2 7DE

Date: 3rd July 2019
Direct line: 0141 314 0213
E-mail: WOSREC6@ggc.scot.nhs.uk

Dear Mrs Marscheider

Study title: An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life
REC reference: 18/WS/0168
Amendment number: Amendment 1 15/04/19 - REC REF AM01
Amendment date: 25 April 2019
IRAS project ID: 245506

The above amendment was reviewed by the Sub-Committee in correspondence.

Summary of Amendment

Change of Chief Investigator from Dr Alison Jackson to Dr Naomi White.

Ethical opinion

The members of the Committee taking part in the review gave a favourable ethical opinion of the amendment on the basis described in the notice of amendment form and supporting documentation.

Approved documents

The documents reviewed and approved at the meeting were:

Document	Version	Date
Notice of Substantial Amendment (non-CTIMP) (Amendment 1 15/04/19)	Amendment 1 15/04/19 - REC REF AM01	25 April 2019
Participant consent form	7	10 April 2019
Participant information sheet (PIS) (Full Version)	8	10 April 2019
Summary CV for Chief Investigator (CI) (Dr Naomi White)		

Membership of the Committee

The members of the Committee who took part in the review are listed on the attached sheet.

Working with NHS Care Organisations

Sponsors should ensure that they notify the R&D office for the relevant NHS care organisation of this amendment in line with the terms detailed in the categorisation email issued by the lead nation for the study.

Statement of compliance

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

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18/NHS/0188:	Please quote this number on all correspondence
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Yours sincerely

Mrs Liz Jamieson
REC Manager
On behalf of Dr Stewart Campbell, Chair
Chair

E-mail: WoGRECS@ggc.scot.nhs.uk

Enclosures: *List of names and professions of members who took part in the review*

West of Scotland REC 5

Attendance at Sub-Committee of the REC meeting on 28 June 2019

Committee Members:

Name	Profession	Present	Notes
Dr Stewart Campbell	Consultant Physician & Gastroenterologist (CHAIR)	Yes	
Canon Matt McManus	Retired Parish Priest (Vice-Chair)	Yes	

Also in attendance:

Name	Position (or reason for attending)
Mrs Liz Jamieson	REC Assistant

Appendix 2.2: NHS Research And Development Approval



Clinical Research & Development
Dykebar Hospital, Ward 11
Grahamston Road
Paisley, PA2 7DE
Scotland, UK

Administrator: Mrs Elaine O'Neill
Telephone Number: 0141 814 4001
E-Mail: elaine.o'neil@ggc.scot.nhs.uk
Website: <http://www.nhs.org.uk/about-us/professional-support-312/research-development/>

21/06/2019

Mrs Evelyn Marscheider
University of Glasgow
1st floor Admin Building
Garthavel Royal Hospital
1055 Great Western Road
Glasgow G12 0XH

NHS GG&C Board Approval

Dear Mrs E Marscheider,

Study Title: An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery
Principal Investigator: Mrs Evelyn Marscheider
GG&C HB site: Royal Hospital of Children - Paediatric Cardiology Dept
Sponsor: NHS Greater Glasgow and Clyde
R&D reference: GN18CA404P
REC reference: 18/WS/0158
Protocol no: V4.1; 19/10/18
(including version and date)

I am pleased to confirm that Greater Glasgow & Clyde Health Board is now able to grant Approval for the above study.

Conditions of Approval

1. For Clinical Trials as defined by the Medicines for Human Use Clinical Trial Regulations, 2004
 - a. During the life span of the study GGHB requires the following information relating to this site:
 - i. Notification of any potential serious breaches.
 - ii. Notification of any regulatory inspections.

It is your responsibility to ensure that all staff involved in the study at this site have the appropriate GCP training according to the GGHB GCP policy (www.nhs.org.uk/content/default.asp?page=51411), evidence of such training to be filed in the site file.

2. For all studies the following information is required during their lifespan.

- a. First study participant should be recruited within 30 days of approval date.
- b. Recruitment Numbers on a monthly basis
- c. Any change to local research team staff should be notified to R&D team
- d. Any amendments – Substantial or Non Substantial
- e. Notification of Trial/study end including final recruitment figures
- f. Final Report & Copies of Publications/Abstracts

Please add this approval to your study file as this letter may be subject to audit and monitoring.

Your personal information will be held on a secure national web-based NHS database.

I wish you every success with this research study

Yours sincerely,

Mrs Elaine O'Neill
Senior Research Administrator

CC: Ms Emma-Jane Gealt (University of Glasgow)

Appendix 2.3: Participant Information Sheets

Version 3, November 2018



Participant Information Brief Summary

An exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

We are carrying out a study to explore what it is like to receive a diagnosis before birth of Congenital Heart Disease (CHD) for your infant and what it is like when your infant undergoes cardiac surgery within their first year of life. We hope the findings from the study might help to improve the experiences of other families in the future.

What would happen if I took part in the research?

Participation would involve a 60-minute interview with Evelyn Marscheider, Trainee Clinical Psychologist. It would involve discussing your experiences of having a child who received a diagnosis of CHD before birth. We would like to hear about your experiences throughout your child's health care experience, for example following diagnosis, prior and during surgery, and admission to the Paediatric Intensive Care Unit. The interviews will be voice recorded, however the information that you share will be anonymised.

Do I have to take part?

No – your participation is voluntary and deciding not to take part will not affect the care you or your child receives in any way. You can withdraw from the study at any point without giving a reason with your and your child's care unaffected.

What to do if I am interested in taking part in the research?

If you might like to take part in the study, please speak to a Cardiac Liaison Nurse who can provide you with additional information. They will provide you with the full Participant Information Sheet which contains a detailed description of the study and what is involved. You would be asked to fill in a participant response form, providing contact preferences and details. Following this Evelyn, the researcher, will contact you to provide further information, answer any questions you might have, and arrange a suitable date and time for the interview.

Thank you for taking the time to read this information summary leaflet.

Version 8, April 2019



Participant Information Sheet – Full Version

An exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

This research project is looking at mothers' experiences of having a child with Congenital Heart Disease (CHD). This information sheet contains the full details of the research study for you to read before you consider whether you would like to take part. Thank you for taking the time to read this information.

Who is carrying out the research?

Evelyn Marscheider, (Trainee Clinical Psychologist) from the University of Glasgow is carrying out the study as part of her qualification for the Doctorate in Clinical Psychology. The project is supervised by Dr Naomi White, University of Glasgow, Institute of Health & Wellbeing, Dr Susan Cassidy, NHS Greater Glasgow & Clyde, and Dr Ruth Pegler, NHS Greater Glasgow & Clyde.

Why is this study being carried out?

We are interested in hearing about mothers' experiences whose infant received a diagnosis before birth of CHD and undergone cardiac surgery within the first year of life. There has been very little research carried out in the UK on this topic. Your experiences may help the hospital to gain a more in-depth understanding of the impact of receiving an antenatal diagnosis of CHD and subsequent health care experiences and may help to improve the services they provide to families.

Why am I being invited to take part?

You are being invited to take part because you have an infant who received a diagnosis of CHD before birth and either has undergone cardiac surgery or is expected to undergo surgery within the first year of their life. This may mean that you have not yet given birth to your infant.

Do I have to take part?

No – your participation is voluntary, and deciding not to take part will not affect the care you or your child receives in any way. You can withdraw from the study at any point without giving a reason with your and your child's care unaffected.

What would happen if I took part in the research?

If you are interested in taking part, please complete a participant response form and pass this on to a member of the cardiology team. Alternatively, please feel free to contact the main researcher directly by email (e.marscheider.1@research.gla.ac.uk). The main researcher will then contact you to answer any questions and arrange an appointment to carry out the interview at a suitable time at Glasgow's Royal Hospital for Children. The researcher is only looking to interview approximately 10 mothers, therefore depending on how many people offer to take part, you may not be invited for interview however the researcher will still be in contact to inform you of this.

What will happen in the research interviews?

Prior to the interview, the main researcher will ask you to sign a consent form to say that you have read and understood the information in this leaflet and that you still wish to take part in the study. You will also be asked to complete a Family Information Form with the main researcher which includes details about yourself but also details about your infant's diagnosis and surgery. The research interview will last for around 60 minutes. It will involve discussing your experiences of having a child who received a diagnosis of CHD. We would like to hear about your experiences throughout your child's health care experience, for example following diagnosis, prior and during surgery, and admission to the Paediatric Intensive Care Unit. You will be asked some specific questions but the researcher would like to hear about the parts of your experience that are important to you. The interviews will be voice recorded and transcribed.

Who will know I am taking part?

The clinicians working with you and your child may know that you are taking part in this study. However, the information that you share during the interviews will be kept confidential and will be anonymised. Confidentiality would only be broken if it was believed that you or someone else was at risk of harm. In this situation, the main researcher will inform you of the appropriate people that would need to be informed.

What will happen to the information I provide?

All study data will be held in accordance with The General Data Protection Regulation (2018) and the Data Protection Act (2018). The voice recording device used will be kept securely at the Royal Hospital for Children or Glasgow University. It will only be accessible to the researchers of this study and the study sponsor, NHS Greater Glasgow and Clyde, if they wish to audit the study. The transcription will be retained until the study is completed, when it will be deleted from the device. The transcript will be stored on a password protected laptop, with the data backed up on a secure network. The results of this study will be written up as a report and submitted as part of the researcher's Doctorate in Clinical Psychology. It will be saved electronically by the University of Glasgow and accessible to future students. The results will be presented to the Cardiology department and it is hoped that the results will be submitted for publication in a scientific journal and presented at conferences. There will be an opportunity for you to be notified of the study findings if this is your preference. It is possible that information that you share about your experiences will be quoted within the report. However, any identifiable information will be removed. Following completion of the study, anonymised research data will be stored at the University of Glasgow site at Gartnavel Royal Hospital. Audio recordings will be deleted on completion of the study and will not be stored long term.

Are there any benefits to taking part?

There are no direct benefits to you in taking part in this study. However, the information that we learn from the study will help to improve the understanding of your experiences. In turn, it is hoped that services can be improved in order to provide the best support for people. In addition, it may be helpful to have an opportunity to talk about the experiences that you have been through.

Are there any downsides to taking part?

The interview may bring up thoughts, feelings or memories that are upsetting for you. At any time, you can have a break or end the interview. If you feel upset after the interview, the researcher will be available to talk with you. A Clinical Psychologist based at the hospital would also be available to provide additional support. You will be provided with information about additional supports that are available.

Who has reviewed the study?

This study has been reviewed by staff within the University of Glasgow's Doctorate in Clinical Psychology team. It will also be reviewed by an NHS Research Ethics Committee and the NHS GG&C Research and Development Department.

What do I do if there is a problem?

If you are unhappy about any aspect of the study, then please speak to the main researcher who will do their best to address your concerns. If this is not satisfactory for you, you are able to complain formally through the NHS complaint system.

Any further questions?

If you have any questions please do not hesitate to contact the main researcher (e.marscheider.1@research.gla.ac.uk) or Dr Naomi White who is supervising this research (naomi.white@glasgow.ac.uk). You may also contact Prof Tom McMillan (thomas.mcmillan@glasgow.ac.uk), a professor at the University of Glasgow who is independent of the research.

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

Appendix 2.4: Participant Consent Forms

Version 4, November 2018



Participant Response Sheet

An exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

If you have read the Participant Information Form (full version) for the research study and are interested in being contacted to take part, or to discuss the study further, then please complete a few details and pass this onto a member of the Cardiology Team or contact me directly.

Your name:

Your child's name:

Email address:

Telephone number:

If you wish to be contacted by telephone, is it ok to leave a voice message if you are not available to take the call? (*Please circle*): Yes No

If you are consenting for further contact about this study after your child's surgery please include the estimated date of your child's surgery below:

.....

Thank you for taking the time to read the information leaflet and considering taking part in this research study.

I look forward to speaking with you soon.

Yours sincerely,

Evelyn Marscheider
Trainee Clinical Psychologist
e.marscheider.1@research.gla.ac.uk

Appendix 2.5: Family Information Form

Version 4, October 2018



Family Information Form

An exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

Research Pseudonym:

Mother

Age:

Number of children:

Employment:

Town/City of residence:

Relationship status:

To be completed by researcher

Infant

Age:

Date of operation:

Diagnosis:

In-patient/out-patient:

Length of hospitalisation:

Length of time in ICU:

Length of time on ward:

Length of time since discharge:

Appendix 2.6: Semi-Structured Interview Schedule

Version 4, October 2018



Interview Schedule

An exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

Introduction

- Introduce self and my role as researcher/trainee clinical psychologist.
- Thank participant for agreeing to take part in the study.
- Remind participant about confidentiality and its limits, as well as data anonymity.
- Remind participant that they can stop for a break at any point in the interview if they need to do so, and that they can withdraw from participation at any point without any impact on the service or care they or their child receives.
- Discuss how I will ask some specific questions to help guide the interview but that I am interested in hearing about their experience from their point of view.
- Check consent and get participant to sign written consent form.

Interview questions:

Time of diagnosis

- Can you tell me about when you first heard that there might be a problem with [infant's name]'s heart?

Treatment plan

- What was your understanding of the type of treatment [infant's name] would need?

Before surgery

- Can you tell me about how the lead up to surgery was?

During surgery

- How did you feel while [infant's name] was in surgery?

After surgery

- What was [infant's name]'s recovery period like after surgery?

Future

- Looking back over your experiences what would you say has been the most difficult part(s)?
- Looking forward, what are you most focused on?

Support

- Can you tell me a bit about what kind of support you received within the hospital?
- How did the level of support that you received compare to your expectations?
- Is there anything that the staff could have done differently for you?

Final questions

- What advice would you give parents whose child has just been diagnosed with CHD or are going through the same experience you have?
- Is there anything I haven't asked you about that you think is important that I should know about your experience?

Examples of general prompts that will be used throughout the interview:

- "Can you tell me a bit more about that?"
- "What was that like for you?"
- "How did it make you feel?"
- "Could you give me an example of that?"
- "What do you mean by...?"
- "What did you think about that?"

Appendix 2.7: Sample Of Analysed Transcript

55	Just get through it	I think you just do. I mean I had [name] my other one [child] so he was, he was a great distraction and I've got a great family and like they just, we just kind of muddled our way through. I don't know, you just do, you just do.
56		
57		
58		Great family support
59		'muddled' - ammass/ not organised/complicated way.
60		Copings a family unit
61		
62	Wife family support journey	Yeah great family support. My mum and dad live in the same sort of area, and my sister as well like we were all devastated but we all just took it each step at a time. I started coming for quite regular scans so you were just living Thursday to Thursday really. And then, I don't know, you just kind of, I don't know, you just get through it. You just find a way.
63		
64	Find a way to get through it	
65		
66		What sticks out most from that period for you?
67		
68	Experience of antenatal discussions	I just kinda always knew that a termination wasn't an option for me. I just would never ever have done it. I just remember like sometimes like kind of having to justify that a little bit and
69		
70	Partners difficulties coping	erm like my husband never coped very well. He went to bits really. He kinda thought about, he was you know discussing a termination and things like that, it was just such a [pause] I felt like I was fighting the babies corner because he was mine and he just, I don't know, I don't know, it was a kind of isolating time. You are kind of on your own and you were trying
71		
72	Experience of a battle fight	to stay strong, I was trying to stay strong for my other boy and [pause] I don't know. Like I say you just live Thursday to Thursday and you just hope that the next Thursday you get a little bit more positive news. We kind of went through a stage where we got terrible news like every week, it was going from bad to worse erm and then it was all very much dependant on a part of this valve growing erm and obviously once you get to a point in pregnancy that the scans they don't see enough. So always very much dependant on whether or not when he was born if this valve was there that they could do anything with it so everything was up in the air. Although we tried to plan, it was so hard to plan because
73	Isolating time	
74		
75		
76	Hope for positive news	
77		
78		
79		
80	Experience of uncertainty of diagnosis	Everything was up in the air

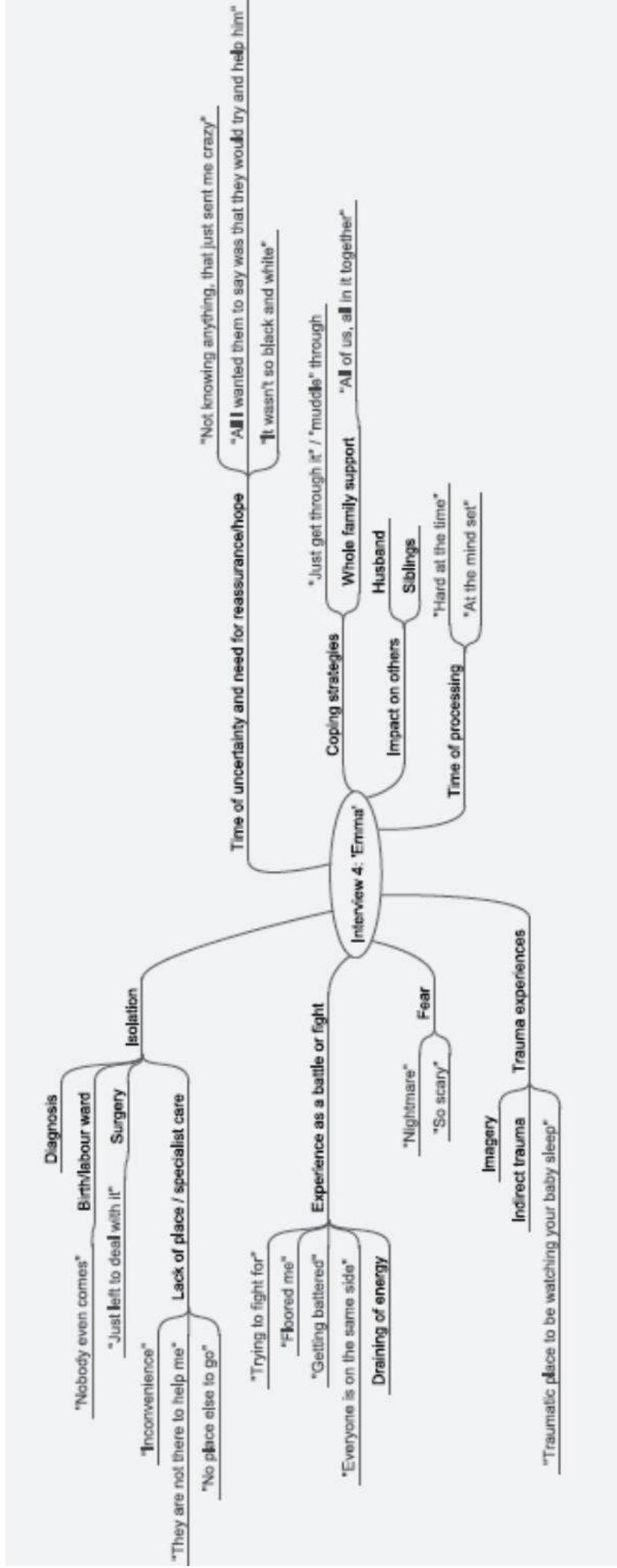
Appendix 2.8: Example Of Emergent Themes and Illustrations For Individual Mother

Emergent Themes for ‘Emma’

<u>Theme / Subthemes</u>	<u>Illustrations</u>
Coping strategies “Just get through it” “Muddled”	<p>“I think you just do [...] we just kind of <u>muddled</u> our way through. I don’t know, you just do, you just do.” (55)</p> <p>“I don’t know, you just kind of, I don’t know you just get through it. You find a way.” (63)</p> <p>“I don’t know, I think you just get through it don’t you. Just <u>muddle</u> through it.” (301)</p>
Whole family support	<p>“Yeah great family support. My mum and dad live in the same sort of area, and my sister as well like we were <u>all</u> devastated but we <u>all</u> just took it each step at a time” (61)</p> <p>“Yeah like my mum came with me everyday [...] my mum would just come sit with me everyday for like company. Or my sister would come.” (325)</p> <p>“They were just as <u>invested</u> in him as I was so they wanted to be there. So right from the scan it was <u>all</u> of us, <u>all</u> in it <u>together</u>.” (334)</p> <p>“I feel so sorry for people who don’t have families” (575)</p>
Support from partner	<p>“It got to a point where I was like you either need to get onboard with this plan or you go away because you are draining me and I don’t have the energy to worry about you and the baby” (342)</p>
Whole experience as a battle / fight / competitive game	<p>“I felt like I was <u>fighting</u> the <u>baby’s</u> corner cause he was mine” (72)</p> <p>“At that point [pause] you are <u>trying to fight for</u>, oh I’m going to get upset here [tears], <u>try to fight for</u> your family life that you’ve got [pause] you know it’s just hard, it’s just hard.” (102)</p> <p>“It <u>floored</u> me because I was not prepared for cerebral palsy [...] so that full week we were just <u>getting</u> <u>battered</u> with you know things that could be wrong.” (168)</p> <p>“Waiting ‘til the meetings that week is just torture” (176)</p>

	<p>“Like that <u>killed</u> me” (348)</p> <p>“Just was like, I felt like I was fighting an up hill battle.” (509)</p> <p>“I think you have to remember that <u>everyone</u> is on the same side. That you can get really frustrated. Everyone wants what is best for your baby but at the you can get a bit, arguing with people because it is not happening quick enough. But you’ve got to remember that <u>everyone</u> is on the <u>same side</u>. Nobody wants anything bad to happen” (623)</p>
Impact of experience on family	<p>“My husband never coped very well. He went to bits really.” 70</p> <p>“He’s always a person that is in control and all of a sudden for this, to not be in any, there was nothing that he could do to help.” (345)</p>
On partner	<p>“I struggled with that as well because I didn’t want [son’s] life to be in a hospital. Like that <u>killed</u> me. Having to bring him to hospital everyday. I still cry about that [...] you know, it’s just so <u>unfair</u> on him, and around all these sick kids [sigh]” (347)</p>
- helplessness	<p>“I feel that did have an effect because he was spoilt at that point you know cause it was just give him anything to just shut him up while the doctor came to talk to you, so I do think it has had a negative impact on his behaviour a wee bit.” (355)</p>
- coping	<p>“Feel like he [baby] gets so much attention that [son] is playing up a wee bit now because he’s wanting, he knows and can understand now what is going on [...] but at the time I just felt so guilty that he was in hospital” (364)</p>
On siblings	<p>“You kind of have a bit of normality [...] but like we had [son] and if you have other kids you just have to because it is not fair on them.” (535)</p>
- guilt	
- impact on behaviour	
- need for normality	

Visualisation of data using 'FreeMind' software



Appendix 2.9: Example Of Emergent Themes and Illustrations For All Mothers

<u>Subtheme</u>	<u>Illustrations</u>	<u>Representations</u>
Nature of the battle	<p>“Someone said it’s like snakes and ladders, which it is, because sometimes you think you are going really well and all of a sudden something comes, and it <u>completely knocks you out.</u>” Jen</p> <p>“<u>Horrendous</u>” Jen, Sophie, Emma</p> <p>“<u>Hit</u>” Jen, Linda / “It’s only now that it is <u>hitting me</u>” Jen</p> <p>“I think it’s really difficult to process it and live it. Erm [pause] and I think that is what’s <u>hitting me now</u>, is that all the aspects of what we have been through” Jen</p> <p>“<u>Nightmare</u>” Linda / “A <u>nightmare</u> really” Emma</p> <p>“<u>Nothing can prepare you</u>” Sophie</p> <p>“<u>Horrific</u>” Sophie</p> <p>“It was probably the <u>worst two weeks in my life</u>” Sophie</p> <p>“<u>Fighting another battle</u>” Sophie</p> <p>“You just <u>live in fear</u> basically” Sophie</p> <p>“I felt like I was <u>fighting the baby’s corner</u>” Emma</p> <p>“<u>Floored me</u>” Emma</p> <p>“<u>Killed me</u>” Emma</p> <p>“<u>Getting battered</u>” Emma</p> <p>“<u>Torture</u>” Emma</p> <p>“Just was like, I felt like I was <u>fighting an uphill battle</u>” Emma</p> <p>“I wouldn’t wish it on my <u>worst enemy</u>” Emma</p> <p>“<u>On edge all the time</u>” Claire</p>	<p>Jen</p> <p>Sophie</p> <p>Emma</p> <p>Linda</p> <p>Claire</p>

Surrounding environment as continuation of battle scene

“Walking in, and seeing the other kids as well, you know, they are really, really sick around you. It’s really difficult you know” Sophie

“lot really just babies, I mean you are watching big kids, some 18 years, I mean some died. Do you know what I mean? They are just pulling curtains, and you know that someone is just like [sigh] some of the sights we seen [...] it’s quite a traumatic place to be watching your baby sleep [...] you were just seeing horrendous sights, families like in bit, it was just terrible.”
Emma

Trauma symptoms

“I had like flashbacks that I tried to imagine myself how the operation would like. And it was all of that imagining you know, the brain was playing with me. It was very, very weird and difficult.” Linda

“Heard an arrest call and I just froze because it [pause] brought me back to that time when Lily, where all the doctors just ran and I knew it was her. And it was the strangest feeling. I’ve never experienced it, as if the ground had swallowed me up and I couldn’t get out, I couldn’t do anything, and all I could her was a buzzer.” Sophie

“But my partner he, he has horrendous nightmares.” Sophie

“You couldn’t sleep because your mind is going a hundred miles an hour” Emma

“Like I have my moments sometimes like where I am upset and sometimes it does, it can kind of come on and overwhelm you, like sometimes, like I’ve maybe not dealt with it at the time.” Claire

Appendix 2.10 COREQ

COREQ (Consolidated criteria for Reporting Qualitative research) Checklist

A checklist of items that should be included in reports of qualitative research. You must report the page number in your manuscript where you consider each of the items listed in this checklist. If you have not included this information, either revise your manuscript accordingly before submitting or note N/A.

Topic	Item No.	Guide Questions/Description	Reported on Page No.
Domain 1: Research team and reflexivity			
<i>Personal characteristics</i>			
Interviewer/facilitator	1	Which author/s conducted the interview or focus group?	39
Credentials	2	What were the researcher's credentials? E.g. PhD, MD	Front cover
Occupation	3	What was their occupation at the time of the study?	41
Gender	4	Was the researcher male or female?	41
Experience and training	5	What experience or training did the researcher have?	41
<i>Relationship with participants</i>			
Relationship established	6	Was a relationship established prior to study commencement?	38
Participant knowledge of the interviewer	7	What did the participants know about the researcher? e.g. personal goals, reasons for doing the research	included in PG
Interviewer characteristics	8	What characteristics were reported about the interviewer/facilitator? e.g. Bias, assumptions, reasons and interests in the research topic	41
Domain 2: Study design			
<i>Theoretical framework</i>			
Methodological orientation and Theory	9	What methodological orientation was stated to underpin the study? e.g. grounded theory, discourse analysis, ethnography, phenomenology, content analysis	37
<i>Participant selection</i>			
Sampling	10	How were participants selected? e.g. purposive, convenience, consecutive, snowball	38
Method of approach	11	How were participants approached? e.g. face-to-face, telephone, mail, email	38
Sample size	12	How many participants were in the study?	38
Non-participation	13	How many people refused to participate or dropped out? Reasons?	38
<i>Setting</i>			
Setting of data collection	14	Where was the data collected? e.g. home, clinic, workplace	39
Presence of non-participants	15	Was anyone else present besides the participants and researchers?	39
Description of sample	16	What are the important characteristics of the sample? e.g. demographic data, date	38-39
<i>Data collection</i>			
Interview guide	17	Were questions, prompts, guides provided by the authors? Was it pilot tested?	40
Repeat interviews	18	Were repeat interviews carried out? If yes, how many?	N/A
Audio/visual recording	19	Did the research use audio or visual recording to collect the data?	40
Field notes	20	Were field notes made during and/or after the interview or focus group?	41
Duration	21	What was the duration of the interviews or focus group?	39-40
Data saturation	22	Was data saturation discussed?	37
Transcripts returned	23	Were transcripts returned to participants for comment and/or	-

Topic	Item No.	Guide Questions/Description	Reported on Page No.
		correction?	
Domain 3: analysis and findings			
<i>Data analysis:</i>			
Number of data coders	24	How many data coders coded the data?	40
Description of the coding tree	25	Did authors provide a description of the coding tree?	40
Derivation of themes	26	Were themes identified in advance or derived from the data?	40
Software	27	What software, if applicable, was used to manage the data?	40
Participant checking	28	Did participants provide feedback on the findings?	
<i>Reporting</i>			
Quotations presented	29	Were participant quotations presented to illustrate the themes/findings? Was each quotation identified? e.g. participant number	42-49
Data and findings consistent	30	Was there consistency between the data presented and the findings?	42-49
Clarity of major themes	31	Were major themes clearly presented in the findings?	42-49
Clarity of minor themes	32	Is there a description of diverse cases or discussion of minor themes?	42-49

Developed from: Tong A, Sainsbury P, Craig J. Consolidated criteria for reporting qualitative research (COREQ): a 32-item checklist for interviews and focus groups. *International Journal for Quality in Health Care*. 2007. Volume 19, Number 6; pp. 349 – 357

Once you have completed this checklist, please save a copy and upload it as part of your submission. DO NOT include this checklist as part of the main manuscript document. It must be uploaded as a separate file.

Appendix 2.11: Major Research Project Proposal

Version 4, October 2018

Major Research Project Proposal

Title: An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery in the first year of life

2292992

Date: 19th October 2018

Version number: 4.1

Abstract

Background

The term “congenital heart disease’ (CHD) refers to abnormalities of the heart that are present from birth, and typically require timely medical intervention. The unique stressors faced by parents whose children have CHD can cause increased psychological distress, especially for mothers. Ongoing stress and mental health difficulties may impact parental quality of life and their capacity for optimal parenting. More research is needed into the lived experiences of mothers whose infants are diagnosed with CHD and undergo surgery to help understand their experiences and target interventions appropriately. In particular, there is limited research exploring mothers’ experiences following an antenatal diagnosis.

Aims

This study aims to explore the lived experiences of mothers whose infants were given an antenatal diagnosis of CHD, and who had undergone surgery within the first year of life.

Methods

Approximately 10 mothers will be recruited through purposive sampling. Their lived experiences will be explored through semi-structured interviews. The interviews will be transcribed and analysed using Interpretative Phenomenological Analysis to explore emerging themes.

Applications

The findings may enhance our clinical understanding of the mothers’ experiences. The in-depth information collected may also inform service development to provide insight into potential service improvements that may help to manage increased psychological distress.

Introduction

Congenital Heart Disease

The term “congenital heart disease’ (CHD) refers to abnormalities of the heart that are present from birth and typically require medical intervention soon after birth and follow-up care for life. Cardiac conditions comprise the greatest proportion of all birth defects, are a major cause of foetal and infant mortality, and are the most common group of structural malformations in children (Lissauer & Clayden, 2011). Recent advances in treatment and survival of infants has led to the transformation of CHD from a terminal to a chronic condition. With increased numbers of children living with CHD there has been a shift in focus from survivorship to improving psychological outcomes and quality of life for children with CHD and their parents.

Experiences of Parents

Having a child with CHD can provide parents with unique challenges and stressors. Compared to parents of healthy children and with other conditions, those whose children had CHD reported greater levels of psychological symptoms including anxiety and depression (Lawoko & Soares, 2002b) as well as anger and hopelessness (Jackson et al., 2015). Such psychological symptoms and distress have been shown to impact on parents throughout the continuum of their child’s health care experience (Soulvie et al., 2012), with incidents of parents experiencing trauma symptoms consistent with an acute stress disorder (Candice Franich-Ray et al., 2013). Parental stress has been found to be significantly higher when their child underwent cardiac surgery during the first year of life than compared to all other ages up to the age of 16 years (Franck et al., 2010). Increased parental stress during their child’s hospitalisation has been associated with poorer psychological outcomes for the child following discharge (Anne E. Kazak et al., 2006). In addition, continued parental stress and mental health difficulties may impact parental quality of life and their capacity for optimal parenting and responsiveness.

Experiences of Mothers

Recent studies have indicated gender differences between the experiences of mothers and fathers (Jackson et al., 2015). Fathers have been found to focus more on containing emotions and focusing on practical tasks (Gower et al., 2017), and experiencing the need to protect their wife as well as their child (Wei et al., 2016). In addition, mothers have been reported to experience a higher level of stress than fathers at time of diagnosis (Bevilacqua et al., 2013a) and prior to their child's heart surgery (E M Utens et al., 2000).

A number of qualitative studies have developed our understanding of mothers' experiences (Bruce et al., 2014; Harvey et al., 2013; S.-F. Lan, Mu, & Hsieh, 2007; Sabzevari et al., 2016). These studies include mothers of children with various CHD diagnoses and at different time points in their child's health care experience. Despite varied focuses, common themes emerge from these studies including reports of feeling a range of intense fluctuating emotions, difficulties with uncertainty and the unknown, difficulties with navigating the medical world, trying to mother through it all, and the importance of support throughout. Critical time points of a mother's experience include time of diagnosis, handing their child over to the surgical team, during surgery, and visiting their child in the paediatric intensive care unit after surgery (Wei et al., 2016).

There are currently very few published studies that explore the experiences of mothers during the course of their infant's antenatal diagnosis of CHD and subsequent surgery and hospital admission. Few studies of this nature have been completed with a British population, and the author is not aware of any published studies to date based in Scotland. It is hoped that the rich information gathered from this research will enhance our understanding of the experiences of this group of mothers. In addition, a recent qualitative study, with 154 parents, identified that during their infant's (0-3 months) hospitalisation for cardiac surgery, parents reported the need for increased quantity and quality of

information and emotional support from hospital staff (Kosta et al., 2015). Therefore, by gathering greater detail into mothers' experiences, the study also hopes to provide insight into potential service improvements that may help to manage increased psychological distress.

Aims

This study aims to explore the lived experiences of mothers whose infants were given an antenatal diagnosis of Chronic Heart Disease, and who have undergone surgery within the first year of life at Glasgow's Royal Hospital for Children.

Plan of Investigation

Design

The study aims to use a retrospective, qualitative design asking participants to discuss recent but previous surgery and admission to Paediatric Intensive Care Unit (PICU), as well as their diagnosis experience. Semi-structured interviews will be conducted with mothers. Interpretative Phenomenological Analysis (IPA; Smith, Flowers & Larkin, 2009) will be employed to analyse the interview transcripts. Given the exploratory nature of this study, IPA was considered to be the most appropriate methodology. IPA allows for a rich, detailed and flexible exploration of an individual's lived experiences and how they make sense of the experience within their wider context (J A Smith, Flowers, & Larkin, 2009). It will therefore allow for unexpected themes to emerge to offer a voice to those immersed in the phenomenon in question.

Participants

The study will aim to recruit 10 mothers whose infant had undergone cardiac surgery prior to 1 year of age at Glasgow's Royal Hospital for Children. Purposive sampling will be used to recruit as mothers will be selected according to specific criteria.

Inclusion and Exclusion Criteria

A main inclusion criterion for this study is the presence of an antenatal diagnosis of CHD. As reported in a review article by (Kolaitis, Meentken, & Utens, 2017a), research findings have been inconsistent with regards to a difference in impact between antenatal and postnatal diagnosis. However, it is agreed that each group of mothers will go through different trajectories based on when a diagnosis was received. Therefore, in order to obtain a homogeneous sample required by IPA, only mothers who received an antenatal diagnosis will be included in this study. Other inclusion criteria will include:

- Mothers with an infant less than 1 year old
- Their infant has undergone at least one surgical procedure due to a major heart defect at Glasgow's Royal Hospital for Children
- The mother is able to communicate their experiences verbally in English

After mothers have completed a response form indicating their interest in completing the study and prior to the researcher approaching them with further information about the study, liaison between the researcher, Cardiac Liaison Nurses, Cardiologists, and Intensivists working with the family will be completed. No identifiable information other than the name of the mother will be exchanged during this consultation. The clinical team will inform the researcher whether or not they feel it is appropriate for the mother to be approached. This will be a simple yes or no. Potential exclusion criteria will include if the infant has a very poor short term prognosis.

Recruitment Procedures

Potential participants will be identified by the clinical team working within the cardiology department at Glasgow's Royal Hospital for Children. An estimated 20 cardiac surgeries are completed per month, with approximately 5 surgeries that would fit the research criteria. A clinician working with the family will initially provide the mother with a participant

information leaflet outlining the purpose of the study and what would happen if they agreed to participate. The leaflet will also include information about confidentiality and the voluntary nature of the study (appendix 4). This will occur either at an antenatal or postnatal stage during a routine clinic or follow up phone call with a Cardiac Liaison Nurse. During clinics the mothers can receive a lot of information therefore it was considered appropriate to receive a shortened version of the participant information sheet initially. Mothers who indicate interest in participating will be given or sent out the full participant information sheet (appendix 5) and asked to complete a response form (appendix 6). The mothers will then be contacted by the main researcher to discuss the study further. The researcher will only contact the mothers following liaison with a clinician working with the family. This is to facilitate discussions regarding whether it would be appropriate for the mother to be included in the study. The decision will consider factors such as the outcome of the surgery, the child's health, and the child's prognosis. Participants will be recruited until the required number of participants has been met.

Research Procedures

Following initial contact with mothers, the researcher will arrange to meet with the mothers individually to conduct the interviews. Semi-structured interviews will be used to explore the participants' experiences, allowing for flexibility of discussions. Written consent will be gained prior to the start of interviews (appendix 7). Participants will also be asked to fill in a demographics form (appendix 8) which will collect basic information regarding the mother and the infant such as age, length of time in ICU, and whether the infant has been discharged from hospital. The interview schedule will be informed by previous research and through consultation with the Cardiology Team at Glasgow's Royal Hospital for Children (appendix 9). The interview will be conducted flexibly following the mothers'

individual narrative, allowing the mothers to develop their own ideas of their experiences. All participants will be asked the same descriptive question at the beginning of the interview, building to questions that are more sensitive in nature. The interview schedule will be piloted on the first three interviews and revisions made if necessary (pilot interviews will be included in the analysis). Due to the potentially distressing nature of the topics, time will be taken to develop a rapport with participants before discussing the potentially challenging topics. The interviews will be recorded and transcribed.

Data Analysis

The transcripts will be analysed in line with IPA methodology (J A Smith et al., 2009). Each transcript will be read on multiple occasions before highlighting and making comments on interesting or significant points. Emergent themes from each interview will then be noted and analysed together before master themes are generated. Approximately three transcripts will be independently analysed by a second researcher, supervising the research, to support that emerging themes adequately captured participants' experiences. A reflective journal will be kept by the main researcher containing perceptions of the data to facilitate the identification and minimisation of bias, to help enrich the developing analysis, and to aid transparency.

Justification of sample size

Studies implementing an IPA approach benefit from an in-depth focus on a small number of participants (J A Smith et al., 2009). The aim will be to recruit 10 mothers. In a recent systematic review of the familial impact of CHD, including qualitative research, studies were excluded if they had a sample size of less than 10 (Jackson et al., 2015). Although the authors did not provide a justification for this exclusion criterion, it is hoped that this research will be used to add to the current literature therefore will aim to meet this

criterion. A sample size of 10 mothers and thus 10 interviews is consistent with Smith et al (2009) recommendation for a professional doctorate.

Settings and Equipment

Interviews will be completed within a private clinic space at the Royal Hospital for Children, Glasgow. This will ensure it is a familiar environment for the participants. An audio recorder will be used to record interviews and a laptop for transcription.

Health and Safety Issues

Research Safety Issues

Interviews will be conducted between normal working hours within the hospital. The researcher will ensure that the field supervisor or another member of the team is contactable during interviews.

Participant Safety Issues

The study will comply with data handling and protection policies in line with the General Data Protection Regulation (2018) and with local NHS policies. Consent and demographic information will be collected using paper documents. During the data collection phase of the study, these questionnaires will be stored at the Royal Hospital for Children in a secure filing cabinet within the Paediatric Clinical Psychology department. Interviews will be recorded on an audio recorder. When not in use, the recorder will be stored in a locked filing cabinet within the Clinical Psychology Department. The voice file will be uploaded to an encrypted laptop with data being backed-up regularly to secure networks. Participants will be given a pseudonym and interviews will be transcribed without identifiers. They will be informed that the interviews will be recorded and all identifiable data will be removed from the transcripts and stored securely. Data will only be accessible to the researchers of

this study and the study sponsor, NHS Greater Glasgow and Clyde, if they wish to audit the study. Following completion of the study, anonymised research data will be stored at the University of Glasgow site at Gartnavel Royal Hospital. It will be deposited with Enlighten for long-term storage. Audio recordings will be deleted on completion of the study and will not be stored long term.

It is recognised that participants may find it distressing to discuss their experiences. If this occurs, time will be taken to manage the distress within and following the interviews by the researcher who is a trainee psychologist experienced in managing psychological distress in a professional capacity and is supervised by a qualified Clinical Psychologist. In addition, an NHS information leaflet for additional support, including the contact details for the Clinical Psychologist linked to the Cardiology Service, will be given to the participants (appendix 10). If there are concerns about risk for the participant or another individual, the researcher will follow local NHS procedures.

Ethical Issues

Ethical approval will be applied for through the Integrated Research Application System from an NHS Research Ethics Committee and management approval from the NHS GG&C Research and Development Department. Informed consent will be obtained from participants prior to the interview ensuring that they understand that they have the right to stop the interview at any point without this affecting the quality of care their infant receives. The researcher will inform the participants of the aims of the study at the initial, informal contact prior to the interviews being arranged.

Financial Issues

The study will require limited funding. Funding will be needed to cover administrative costs such as stationary and printing which is granted by the University of Glasgow, Institute of

Health and Well-Being. Equipment needed for the study can be borrowed from the University of Glasgow. This research will be submitted as part of the main researcher's doctorate in clinical psychology. The main researcher is funded by NHS Education for Scotland.

Dissemination

The research will be formally written up as a submission according to the requirements for the degree of Doctor of Clinical Psychology (DClinPsy) at the University of Glasgow. The results will be presented to the Cardiology department and it is hoped that the results will be submitted for publication in a scientific journal and presented at conferences. All participants will be given the option of receiving a summary of the results of the research. Those that opt in will be sent a summary through liaison with the cardiac liaison nurses after the research has been fully completed.

Proposed Timetable

January 2018:	Proposal submission to Glasgow University for approval
June 2018:	Apply for ethical approval
August 2018:	Recruitment
February 2019:	Transcribing and coding data
March 2019:	Analysis
April-May 2019:	Write up
July 2019:	Submit to Glasgow University

Practical Applications

The findings from this study are expected to enhance understanding of the experiences of this group of mothers. It is hoped that this information will be used to inform clinical

practice and provide a rationale for further research and clinical intervention. The in-depth information collected can inform service development, providing insight into potential service improvements that may help to manage increased psychological distress for this population.

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Appendix 1: Health and Safety Form

WEST OF SCOTLAND/ UNIVERSITY OF GLASGOW DOCTORATE IN CLINICAL PSYCHOLOGY

HEALTH AND SAFETY FOR RESEARCHERS

1. Title of Project	An IPA exploration of the experiences of mothers of infants with congenital heart disease who have undergone surgery.
2. Trainee	
3. University Supervisor	Dr Alison Jackson
4. Other Supervisor(s)	
5. Local Lead Clinician	Dr Susan Cassidy
6. Participants: (age, group or sub-group, pre- or post-treatment, etc)	Mothers whose infants have an antenatal diagnosis of congenital heart disease and have undergone surgery within their first year of life.
7. Procedures to be applied (eg, questionnaire, interview, etc)	Qualitative interviews will be conducted using semi-structured interviews.
8. Setting (where will procedures be carried out?) i) Details of all settings	Interviews will be completed within a private, clinic space at the Royal Hospital for Children, Glasgow. This will ensure it is a familiar environment for the participants. The interviews will take place between normal working hours within the hospital.
ii) Are home visits involved	Y/ <u>N</u>
9. Potential Risk Factors Considered (for	Participants: This participant sample is not normally

<p>researcher and participant safety):</p> <p>i) Participants</p> <p>ii) Procedures</p> <p>iii) Settings</p>	<p>associated with dangerous or unpredictable behaviour</p> <p>Procedures: The procedures in the study are similar to those used by clinical psychologists with these participants and are not normally associated with production of significant distress. However, it is recognised that participants may find it distressing to discuss their experiences.</p> <p>Settings: Meetings and interviews will be conducted within the Royal Hospital for Children where participants routinely attend as in- and out-patients. As a NHS location, there are appropriate health and safety policies and procedures.</p>
<p>10. Actions to minimise risk (refer to 9)</p> <p>i) Participants</p> <p>ii) Procedures</p> <p>iii) Settings</p>	<p>Participants: Mothers of infants that receive an antenatal diagnosis of CHD are able to access clinical psychology services and do not normally pose any risk to the clinician.</p> <p>Procedures: Prior to interviews the researcher will inform the participants of the aims of the study and gain informed consent.</p> <p>If a participant becomes distressed during the interview the researcher will manage this at the time e.g. suggest a short break or end the interview if appropriate. An information sheet of services for additional support will be provided to all participants including the contact details of the departmental Clinical Psychologist.</p> <p>Mothers have routine access to support through the team Clinical Psychologist therefore if concerns are identified, information can be passed on.</p> <p>If a participant discloses information that may indicate risk of harm to themselves or others, the researcher will take action including risk assessment and management, and pass on this information to the appropriate services in line with local NHS and professional policies.</p> <p>Settings: When arranging the interviews, the researcher will ensure that there are local health and safety protocols in place. The researcher will ensure that a qualified member of clinical psychology NHS staff (field supervisor or equivalent) is available to be contacted at times when interviews are being conducted.</p>

Trainee signature: Date:

University supervisor signature: Date:

Appendix 2: Research and Equipment Costs

RESEARCH EQUIPMENT, CONSUMABLES AND EXPENSES

Trainee

Year of Course ...2nd Year..... Intake Year...2016.....

Please refer to latest stationary costs list (available from student support team)

Item	Details and Amount Required	Cost or Specify if to Request to Borrow from Department
Stationary	N/A	N/A Subtotal: -
Postage	N/A	N/A Subtotal: -
Photocopying and Laser Printing	Printing and photocopying: 170 sheets (30 x 3-page participant information sheet; 30 x 1-page response form; 20 x 1-page consent forms; 10 x 3-page interview guides)	Paper: £2.18 Printing/Photocopy: £0.05 x 170 = £8.50 Subtotal: £10.68
Equipment and Software	Digital recorder, laptop, and pedal for transcription	All to be borrowed from department if available Subtotal: -
Measures	N/A	N/A Subtotal: -
Miscellaneous	N/A	N/A Subtotal: -
Total		£10.68

For any request over £200 please provide further justification for all items that contribute to a high total cost estimate. Please also provide justification if costing for an honorarium:

Trainee Signature..... Date.....

Supervisor's Signature Date

Appendix 3: Plain English Summary (word count: 499)

Exploring the experiences of mothers whose infants have congenital heart disease and have undergone surgery

Background

Congenital Heart Disease (CHD) is a general term for a range of birth defects of the heart. For moderate to severe cases, infants frequently require high dependency care with surgery soon after birth. Improvements in medical treatment have led to more children living with CHD. As a result, focus has moved towards exploring the quality of life of children with CHD and their parents. Research suggests that parents, especially mothers, of children who have CHD experience increased levels of distress (Jackson et al., 2015). Such distress is higher if the child requires surgery during their first year of life (Franck et al., 2010). It is important to understand the mothers' experiences during this time. With a better understanding, services may be improved to help manage increased levels of distress.

Aim

This study aims to explore the experiences of mothers whose infants had an antenatal diagnosis of CHD, and had surgery within their first year of life.

Methods

Approximately 10 mothers will be recruited from Glasgow's Royal Hospital for Children. Clinicians working with the mother will help to identify which mothers would be suitable to be contacted, considering aspects such as whether their infant is medically stable. They will provide information about the purpose of the study and what would happen if they agreed to take part. The researcher will then approach mothers who are interested and arrange to meet in a private clinic room at the hospital to complete the interview. Mothers will have time to ask any questions that they have before the interview. The interviews will focus on a broad range of topics and time-points, and will be flexible to each mother's

individual experiences. They will be recorded and qualitatively analysed using Interpretative Phenomenological Analysis to identify and explore themes that the mothers report.

Ethical Issues

Informed consent will be gained from participants prior to the interview. Information shared by participants will be confidential and anonymised. Confidentiality will only be broken if the researcher feels the participant or someone else is at risk. Time will be taken to manage any distress that the participants may experience. Participants will be given information with the contact details of extra supports they can access.

Practical Applications and Dissemination

It is hoped that the study will increase our understanding of the mothers' experiences and highlight service improvements that may reduce levels of distress. Improvements could include increasing support available to the families. The study will be presented to the Cardiology team at Glasgow's Royal Hospital for Children and may be published in a peer-reviewed journal so that findings may inform wider service development.

References

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