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Anker-Petersen, Sonia (2014) *Caregivers' experiences of caring for a child with cardiac arrhythmia who has an automatic external defibrillator: an exploratory study using interpretative phenomenological analysis and clinical research portfolio*. D Clin Psy thesis.

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Caregivers' Experiences of Caring for a Child with Cardiac Arrhythmia who has an
Automatic External Defibrillator: An Exploratory Study using Interpretative
Phenomenological Analysis
And Clinical Research Portfolio

Volume I

(Volume II bound separately)

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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

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October 2014

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Acknowledgements

I would like to thank the caregivers who participated in my research study. It would not have been possible without their input.

I would like to give a huge thank you to my supervisors Dr Sarah Wilson and Dr Kathleen McHugh for their guidance, support and expertise throughout the research process. Thanks to Dr Kenneth Mullen for his advice along the way. I am also grateful to Dr Karen McLeod and Sister Eileen Fern at RHSC, Yorkhill Hospital for their help with the recruitment process.

Finally, I would like to thank my family, boyfriend, friends and fellow trainees for being a huge source of support and encouragement throughout this doctorate. I could not have completed it without having such an amazing support network around me.

Chapter 1: Systematic Review

Parental Experiences of Caring for a Child with Chronic Illness: A Meta-Ethnography

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Abstract

The present study aims to systematically review, critically appraise, and synthesise recent qualitative research on parental experiences of caring for a child with chronic illness. Quality appraisal of the 13 studies identified from the systematic review led to six studies being included in the final synthesis. Meta-ethnography was used to synthesise the studies, leading to six new super-ordinate themes being developed: 1) Emotions 2) Impact on Family Functioning 3) Internal Coping Strategies 4) Co-Parenting 5) External Support and 6) Helping the Child to Cope. The findings and implications for clinical practice are discussed.

Introduction

There is wide variance in the prevalence estimates of childhood chronic illness, with estimates ranging from 0.22% to 44% depending on the definitions and measurement methods used (*cf.* van der Lee, Mokkink, Grootenhuis et al., 2007). Despite this variability, there was clear consensus in the literature that the prevalence of childhood chronic illness had increased over the previous two decades (van der Lee et al., 2007). This was due to increased efficacy of treatments and health care for life-threatening paediatric conditions, and the subsequent increased survival rates of the children affected (van der Lee et al., 2007). Mortality has often been replaced by lifelong morbidity and chronic illness (Mokkink, van der Lee, Grootenhuis et al., 2008).

Due to the broad variability in definitions used in the literature, a definition of chronic conditions in childhood was agreed via a national consensus procedure in 2008 by Mokkink et al. They reached the following definition: “A disease or condition is considered to be a chronic condition in childhood if: (1) it occurs in children aged 0 up to 18 years; (2) its diagnosis is based on medical scientific knowledge and can be established using reproducible and valid methods and instruments according to the professionals; (3) it is not (yet) curable or, for mental health conditions, if it is highly resistant to treatment and (4) it has been present for longer than three months, if it will, very probably, last longer than three months or if it has occurred three times or more during the past year and will probably recur again” (pg. 1446).

Chronic illness can have a significant impact on the affected child, siblings and parents (Cousino & Hazen, 2013). Research indicates there are many stressors which parents experience when caring for a child with chronic illness. These include: financial stress, role strains within the family unit, marital separations, adjustment to working with the medical system, interruptions in daily routines and future plans, and the general uncertainty with regard to the child's prognosis (Brown, Wiener & Kupst et al., 2008). Experiencing these stressors has been found to lead directly and indirectly to anxiety, depression, post-traumatic stress, hopelessness, and feelings of loss of control (Brown et al., 2008).

A random effects meta-analysis carried out as part of a systematic review by Cousino and Hazen (2013, pg.819) indicated that parents of children with chronic illness reported greater general parenting stress than parents of healthy children. In general, the literature suggests a reciprocal relationship between chronic illness and parental adaptation, whereby the child's illness impacts on parents' functioning and parental functioning impacts on the child's adaptation (Brown et al., 2008). Thus, parental experience and adaptation is an important area to research further given the potential lifelong effects on their child's subsequent development and adaptation.

Alongside this there is a growing consensus that the needs, preferences and experiences of service users should be considered in the development and evaluation of service delivery models (Ring, Ritchie, Mandava et al., 2010). In line with this, it is important to gain a deeper level of understanding of what it is like to parent a child with chronic illness, in order to inform service and health care provision for this population. Qualitative research aims to "provide an in-depth understanding of people's experiences, perspectives and histories in the context of their personal circumstances or settings" (Spencer, Ritchie, Lewis et al., 2003, pg. 3), and thus is well-placed to provide deeper insights into phenomena such as parenting a child with chronic illness. A systematic review of the qualitative literature on parental experiences of caring for a child with chronic illness will give an even deeper level of insight into this area of experience.

A metasynthesis carried out by Coffey in 2006 examined 11 qualitative and mixed-method studies, published between 1989 and 2000, of the experience of parents caring for a child with chronic illness. Coffey (2006) found several common themes. She found that parents shared common feelings of grief and fear around the diagnosis and management of the child's illness, and that there was a clear need for support in the early stages. Additionally she found that themes of exhaustion, constant worry, and carrying a burden showed up repeatedly in all of the studies. Depression with suicidal ideation was also present for some parents (Coffey, 2006). Despite the contribution of Coffey's metasynthesis in enriching our understanding of parenting a child with chronic illness, she did not critically appraise the quality of any of the primary qualitative studies included in her metasynthesis. There is a growing argument in the field of qualitative research that policy, practice and clinical decisions developed on the basis of low quality or methodologically flawed research studies risk being flawed in themselves (Dixon-Woods et al., 2004; Walsh & Downe, 2006), and thus the utility of Coffey's metasynthesis to clinicians and researchers can be challenged on this basis.

Additionally, given that paediatric medical treatment and health care will have evolved over the last 10 years, it seems important to consider if this has influenced parents' experiences of caring for a child with chronic illness. The present study therefore aims to systematically review, and critically appraise, the more recent qualitative research literature on parental experiences of caring for a child with chronic illness, in order to further develop our understanding of this important experience based on high quality, methodologically sound research studies.

Aim

The aim of the present study is to explore parental experiences of caring for a child with chronic illness by systematically reviewing, appraising and synthesising published qualitative studies in this area.

Review Question

What is the experience for parents of caring for a child with chronic illness?

Method

Search Strategy

Ovid Medline (R) 1946 to Present was used to search EMBASE and MEDLINE databases and EBSCO host was used to search CINAHL and PsychINFO databases.

Search terms

A broad search strategy using free text was employed due to the well-known difficulty in retrieving qualitative studies through electronic searches (Centre for Research and Dissemination, 2008). All searches were completed using the following terms:

1. Disability OR chronic disease OR chronic illness OR childhood disease
AND
2. Family functioning OR family OR psychological aspect OR family life OR child parent relation* OR adaptive behaviour OR family health OR stress OR depression OR mental stress OR parent*
AND
3. Qualitative research OR experience* OR focus group* OR narrative OR content analysis OR grounded theory OR thematic analysis OR interpretative phenomenological analysis

Studies identified by the electronic search were then sorted using the inclusion and exclusion criteria. The reference lists from the selected studies were also searched for any relevant articles that had not been identified through the electronic search.

Inclusion Criteria:

- Studies that explore the experience of parenting a child with chronic illness
- Studies that utilise a qualitative research design
- The child with chronic illness is between 0 and 18 years old
- The studies are published in English
- The studies are published between 2000 and 2014

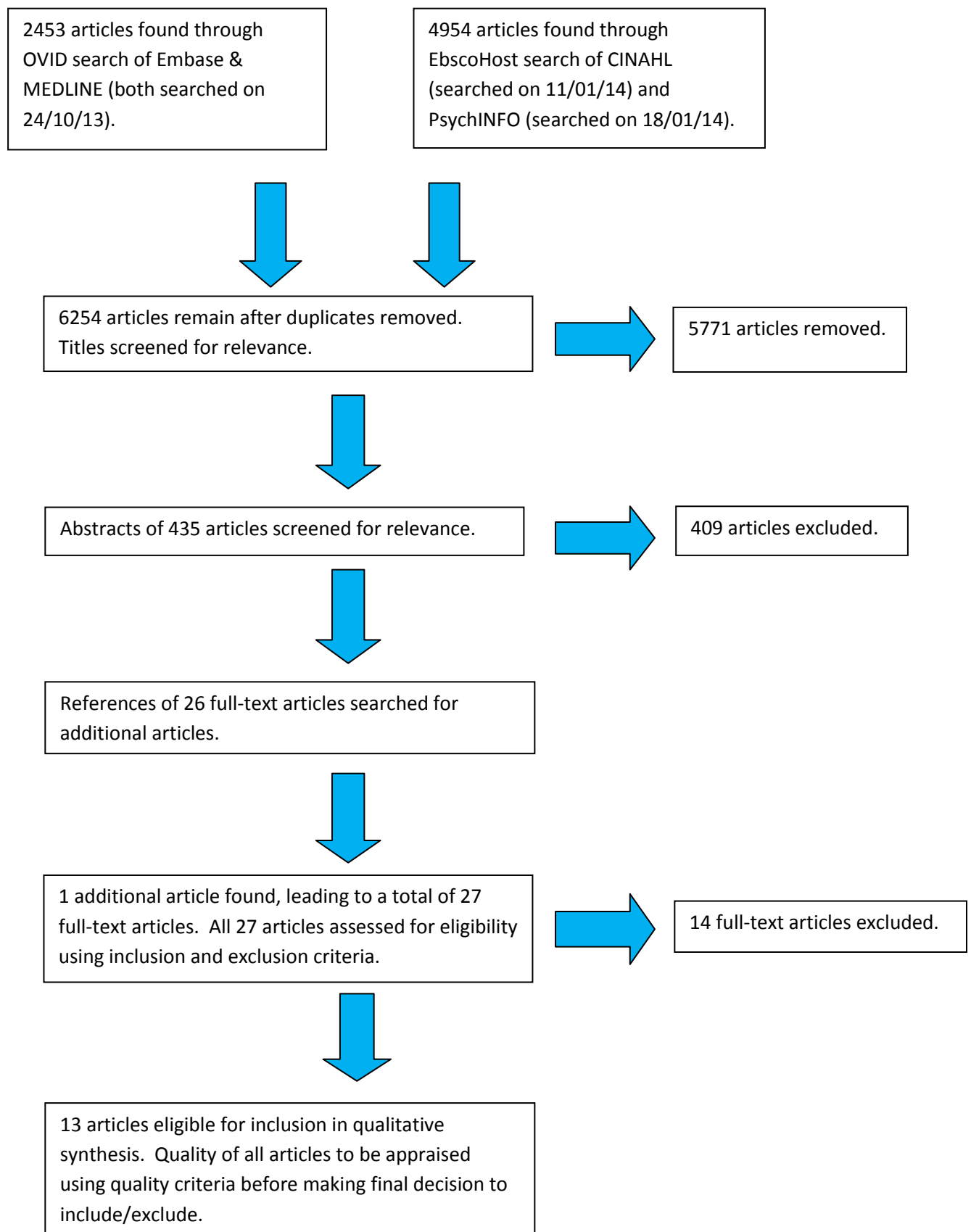
Exclusion Criteria:

- Quantitative studies or mixed-method studies
- Studies focusing on neurodegenerative diseases
- Studies focusing on childhood mental illness
- Studies that are not published in English
- Studies published prior to 2000

Results of search strategy

Figure 1 shows the process and results of the systematic search.

Figure 1. Flowchart of Systematic Search Strategy



Quality Appraisal

The necessity of using quality rating frameworks to rate qualitative research is still subject to debate amongst researchers and clinicians, with some arguing that it is not relevant or helpful (Centre for Research and Dissemination, 2008). In line with many researchers and clinicians in the healthcare field however, it is the author's opinion that high-quality research is essential if it is to be used to inform health-care interventions or to improve people's quality of care (Walsh & Downe, 2006). Walsh and Downe (2006) reviewed, appraised and synthesised existing qualitative research quality assessment frameworks, and developed a comprehensive and practical checklist of criteria that they consider essential to ensure high quality qualitative research. The 12 essential criteria developed by Walsh & Downe (2006) (see Appendix 2) were used to evaluate the methodological quality of each of the 13 studies in this systematic review. The criteria they suggest cover the broad areas of: scope and purpose, design, sampling strategy, analysis, interpretation, reflexivity, ethical dimensions, and relevance and transferability.

Rating the quality of the studies was a challenging task, because even though the guidelines provided by Walsh & Downe (2006) are explicit in what criteria should be met, the extent to which all aspects of these criteria should be present in order for the criteria to be considered 'met' is not explicitly defined. Additionally, judging the quality and integrity of the analytic approaches used in the qualitative studies proved difficult: In order to achieve this task, the author had to familiarise herself with the various approaches and techniques employed in the studies, which was time-consuming. A second researcher independently rated a sample of the studies using the same quality criteria to assess reliability of the primary rater. The overall level of agreement was high at 90%, and any disagreements were resolved through discussion. Overall, it was agreed that the framework proposed by Walsh and Downe is a relatively objective, helpful and meaningful quality framework to use.

Out of the 13 studies, only three met all 12 essential quality criteria. This was a surprisingly low number, but it possibly reflects the lack of guidance that has existed until fairly recently regarding what constitutes quality in qualitative research. A further three articles met 11 out of the 12 essential criteria, and they all failed to meet the criterion for researcher reflexivity. Walsh and Downe (2006) emphasise that

researcher reflexivity is central to qualitative research, however is often discarded from the original research paper in order to meet journal publication word limits. A decision was therefore reached to include these three articles due to them containing rich data which would substantially contribute to the synthesis. Nevertheless, this finding stresses the importance of encouraging journals to allow an increased word limit for original qualitative research articles, so that the integrity of the research can remain transparent after publication.

Table 1 presents an overview of the six studies included and any essential criteria they did not meet. Appendix 3 provides an overview of the seven studies excluded and criteria they did not meet.

Table 1: Quality Appraisal of Papers

Authors (Year)	Country	Method	Quality Rating	Criteria not met	Include in Synthesis?
O'Brien (2001)	USA	Interpretive Interactionism	11/12	No evidence of researcher reflexivity	Yes
Sallfors & Hallberg (2003)	Sweden	Grounded Theory	11/12	No evidence of researcher reflexivity	Yes
McNeill (2004)	Canada	Grounded Theory	12/12	None	Yes
Sullivan- Bolyai, Rosenberg & Bayard (2006)	USA	Fundamental qualitative description	11/12	No evidence of researcher reflexivity	Yes
Tong et al. (2010)	Australia	Thematic Analysis	12/12	None	Yes
Rouf, White & Evans (2012)	United Kingdom	Interpretative Phenomenological Analysis	12/12	None	Yes

Method of Synthesis

Meta-ethnography (Noblitt & Hare, 1988) was used to synthesise the research studies. This approach allows for the synthesis of studies that utilise a variety of qualitative research methods (Ring et al., 2010), and therefore was deemed appropriate for the purpose of the current synthesis. According to Noblitt and Hare (1988), a meta-ethnography “seeks to go beyond single accounts to reveal the analogies between the accounts. It reduces the accounts while preserving the sense of the account through the selection of key metaphors and organizers. The ‘senses’ of different accounts are then translated into one another. The analogies revealed in these translations are the form of the meta-ethnography synthesis” (pg. 13).

Noblitt and Hare (1988) propose that carrying out a meta-ethnography is an iterative process that consists of a series of overlapping phases. Please see Table 2 for an overview of these. These phases were followed when conducting the current meta-ethnography.

Table 2: Seven Phases of Meta-ethnography proposed by Noblitt & Hare (1988)

Phase 1	Getting started- developing a research question.
Phase 2	Deciding what is relevant to the initial interest- deciding on the scope of the synthesis and defining inclusion and exclusion criteria.
Phase 3	Reading the studies- repeated reading of the studies and noting of interpretative metaphors.
Phase 4	Determining how the studies are related by creating a list of key themes from each study and juxtaposing them.
Phase 5	Translating the studies into one another- compare themes and their interactions in one study with themes and their interactions in the other studies.
Phase 6	Synthesising translations by translating studies and their interpretations into each other, leading to a third-order interpretation of studies.
Phase 7	Expressing the synthesis through the most appropriate means.

Results

Table 3 displays the themes that were presented by the authors of the six studies.

Table 3. Themes identified by the authors of the studies included in the meta-ethnography

Author (Year) and Title	Themes
O'Brien (2001), "Living in a house of cards: Family experiences with long-term childhood technology dependence"	<ul style="list-style-type: none"> • Living in a house of cards <ul style="list-style-type: none"> - Vigilance - Advocacy - Reframing • Making sense of life <ul style="list-style-type: none"> - Reconciling the past and present - Changing priorities - Imagining the future • Managing daily life with technology <ul style="list-style-type: none"> - Child's care needs - Time management - Home environment • Maintaining a functioning family <ul style="list-style-type: none"> - Meeting the needs of family members - Finding time for the family - Financial considerations - Maintaining connections with extended family and friends
Sallfors & Hallberg (2003), "A parental perspective on living with a chronically ill child: A qualitative study"	<ul style="list-style-type: none"> • Parental Vigilance <ul style="list-style-type: none"> - Anxiety - Parental protection - Watchfulness • Emotional challenges <ul style="list-style-type: none"> - Uncertainties in parenting - Communication with others - The unknown • Continual adjustment <ul style="list-style-type: none"> - Living here and now - Looking for information - Striving for relief and strength

Author (Year) and Title	Themes
McNeill (2004), "Fathers' experiences of parenting a child with juvenile rheumatoid arthritis"	<ul style="list-style-type: none"> • Diagnosis and Implications of the illness • Emotional responses, self-support and identity as a protector • Fathers' responses: Search for positive meanings • JRA: catalyst for increased involvement and more meaningful relationship • Holistic identity as a father
Sullivan-Bolyai et al (2006), "Fathers' reflections on parenting young children with type 1 diabetes"	<ul style="list-style-type: none"> • Sadness <ul style="list-style-type: none"> - "Shock and awe": the diagnosis - "Suck it up and do it": learning diabetes care - "Staying in the loop": tasks and responsibilities - Partnerships in care: "co-parenting" - Active participation: "staying involved" - Mantra for living with diabetes: child first, diabetes second
Tong et al (2010), "Parental perspectives on caring for a child with chronic kidney disease: an in-depth interview study"	<ul style="list-style-type: none"> • Absorbing the clinical environment <ul style="list-style-type: none"> - confronting the diagnosis - invasive procedures - conflict and trust - varying quality of care - losing ownership - jeopardizing relationship with staff • Medicalizing Parenting <ul style="list-style-type: none"> - A consuming routine - Pressure and isolation - Struggle with feeding - Medical management - Psychological trauma • Disrupting family norms <ul style="list-style-type: none"> - Spousal tension and dependency - Sibling neglect - Household and financial stress - Decision to donate - Social restriction - Avoiding the risk of recurrence • Coping strategies and support structures <ul style="list-style-type: none"> - Internal coping strategies

Author (Year) and Title	Themes
Tong et al (2010) “Parental perspectives on caring for a child with chronic kidney disease: an in-depth interview study”	(Continued): <ul style="list-style-type: none"> - External coping strategies - Information needs
Rouf et al (2012), “A qualitative investigation into the maternal experience of having a young child with severe food allergy”	<ul style="list-style-type: none"> • Parental adjustment- re-establishing an “even keel” <ul style="list-style-type: none"> - Process of emotional adjustment - Practical - Developmental • It’s a big responsibility: living with risk <ul style="list-style-type: none"> - Responsibility and control - Trusting vs. not trusting others - Teaching the child - Managing risk and staying safe • Identity and the social aspects of allergy <ul style="list-style-type: none"> - Not being defined by the allergy and trying to stay normal - Balancing child’s safety with need for social inclusion - Positive and negative support experiences - Social discomfort of allergy

The meta-ethnography resulted in six new super-ordinate themes being developed from the synthesis of these six studies; these themes taken together present a line of argument synthesis, as described by Noblitt and Hare (1988), which arises from a reinterpretation of the previous interpretations. The themes are labelled 1) Emotions 2) Impact on Family Functioning 3) Internal Coping Strategies 4) Co-Parenting 5) External Support and 6) Helping the Child to Cope. Each of these themes will be discussed in turn. All quotations from study participants are presented in italics.

Emotions

Participants in all six studies discussed the emotions that were prevalent for them at various stages of their experience. Several study participants described their

emotions directly following the diagnosis of their child's chronic illness (Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006; Tong et al., 2010; Rouf et al., 2012). The main emotions identified directly following diagnosis involved feelings of shock, devastation, loss, grief, despair, anxiety, fear and sadness. One father whose child was diagnosed with Juvenile Rheumatoid Arthritis (JRA) described searching for meaning and trying to understand why it had happened to him; a phenomenon also reported by several other parents:

"The hardest part is the initial diagnosis. It's the first couple of days. No question about that. You're devastated, you're asking "why me" or "what the hell's going on in life? She's eighteen months old- she's a baby. What's it all about?" So, it's a tough...it's ah, catastrophic, it really is." (McNeill, 2004, pg. 532).

A mother of a child with a nut allergy articulated the feelings of loss she experienced upon hearing the diagnosis:

"Initially just all you see is, you know, all the things that you're not going to be able to do with your child and it's very upsetting." (Julia, 7: 674) (Rouf et al., 2012, pg. 54).

A sense of loss following diagnosis, as conveyed in the above quotation, was reported by parents in most of the studies. Other emotions that occurred immediately following diagnosis, but were less often reported by parents in the studies, included feelings of anger towards the child, depression, and relief at finally knowing what was wrong with their child.

Emotions that were experienced continuously in the day-to-day lives of the parents were also reported in several studies. The most common emotions reported in this context included anxiety, frustration, powerlessness, hope, despair, and sadness. Feelings of powerlessness and helplessness were especially salient in some studies (Sallfors & Hallberg, 2003; McNeill, 2004; Tong et al., 2010), and this appeared to be partly linked to not being able to soothe their child when he/she was in pain. This is emphasised well by a father who was the primary caregiver for his daughter with JRA:

I guess that's the hardest part, dealing with the pain and suffering, just tremendous suffering. And when she says, "Dad take this pain away", I just can't, you know, and dealing with the disappointment- that I just want this pain to go away, I want this JRA, I want this arthritis to go away and the feeling of helplessness [I have]. It is a very disheartening thing. When she cries, there's nothing you can really do because you can massage, you can put hot packs or cold packs, put just about anything, and it's not going to go away. It's just serious, serious pain and that just tears the guts out of you." (McNeill, 2004, pg. 532).

The lack of control over their child's illness was found in other studies to lead to high levels of anxiety for the parents (O'Brien, 2001; Rouf et al., 2012). One mother of a girl with multiple technological needs powerfully conveyed the fragility and uncertainty of their daily life and the anxiety that she felt as a result, despite attempts to be positive:

"All in all, I think we have done well. Our daughter has certainly done well. We have survived. I would like to think that we are more settled now, and in some regards I believe that to be true. But I am not so unrealistic as to not know that everything hangs together by a thread. And as we try to build stability into our future, we still have, we always have, that component of everything hanging together by a thread. And it's like living in a house of cards. It does not take much and it all crumbles." (O'Brien, 2001, pg. 16).

Worrying about the future, particularly the child's ability to manage his/her own condition and his/her future relationships, was a phenomenon reported in four of the studies (O'Brien, 2001; Sallfors & Hallberg, 2003; Sullivan-Bollyai et al., 2006; Rouf et al., 2012). Worry regarding future management was well portrayed by one mother of a boy with a nut allergy:

"I do worry about him being in adolescence (...) I worry that he'll be too cool to carry his EpiPens (...) that he'll have dares with his mates to eat certain things (...) and I need to try to pull back from it because he's only four and we've got a long way to go." (Elizabeth, 6: 124) (Rouf et al., 2012, pg. 55).

This quotation demonstrates that parents' worry can lead to a 'snowballing' effect: they start to worry about potential scenarios that may occur in several years' time, which leads to other worries related to these fictional scenarios. These fictional scenarios provoke real anxiety for the parents despite it being uncertain if they ever will occur in reality.

Parents' worries regarding their child's future relationships were well captured by the following thoughts of a parent of a boy with Juvenile Chronic Arthritis (JCA):

"What will it be like to grow up? Will he be able to have a girl friend? He's so sick, how can he ever meet a girl?" (Sallfors & Hallberg, 2003, pg. 199).

Impact on Family Functioning

The impact on family functioning of caring for a child with chronic illness was described by parents in five of the studies (O'Brien, 2001; Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006; Tong et al., 2010), however, parents' reports of the way in which it had impacted their family varied. The majority of parents' reports in two of the studies (O'Brien, 2001; Tong et al., 2010) described the negative impact on the marital relationship, and their decision to not have any more children as a result of the difficulties they experienced with their current caregiving responsibilities. One mother of a child with chronic kidney disease (CKD) articulated the way in which lack of time, finances and control over their situation led to multiple fights between her and her husband:

"We used to fight a lot because my husband was frustrated that our child wasn't getting better. We were spending all this money, time and energy on getting him well, and nothing was working. We used to fight and he often blamed me for things and I used to fight and say, how can you blame me for it, it's out of my control." (Tong et al., 2010, pg. 554).

Another mother described the significant negative impact of her caregiving role on the marital relationship, and the sense of helplessness she felt in relation to this:

“It’s taken a toll on, to be honest, with dad and I in our relationship. We basically have very little relationship anymore because I don’t have the energy to have a relationship. I don’t have the time anymore. And it’s terrible. I mean, we’ve both lost out”. (O’Brien, 2001, pg. 19).

This mother was the main caregiver for their child, and as a result of her caregiving duties she did not feel that she had any left-over resources to devote to her relationship with the father. This may potentially explain the difference in perspective from parental reports in the other studies, which were generally more positive about the impact of the child’s chronic illness on family functioning. Parents in the other studies often appeared to work as a team and considered themselves to be ‘co-caregivers’ as opposed to there being one main caregiver. This will be discussed further under the theme of ‘Co-Parenting’. Additionally, the nature of the chronic illness may potentially have influenced the way in which it affected family functioning. Negative reports mainly came from parents of children with CKD or multiple technological needs, whereas positive reports mainly came from parents of children with JRA, JCA and diabetes. There were, however, positive and negative reports from parents in all studies, so individual variation in perception also appears to be a factor.

Neglect of healthy siblings was another theme raised by several parents in the studies by O’Brien (2001) and Tong et al (2010). One mother of a child with CKD described this well:

“I had this child who was sick so basically my three year old daughter ended up being put aside. She thought she had the duty of being this perfect child for me. It’s made her life pretty much a nightmare.” (Tong et al., 2010, pg. 554).

As previously mentioned, other parents reported a more positive influence on family functioning (O’Brien, 2001; Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006). This is illustrated by a father of a child with JRA :

I think it can strengthen everything, it can strengthen marriages, it can strengthen families...because you get a better perspective on what's really important in life. You really do.” (McNeill, 2004, pg. 537).

Many parents attributed the positive impact on family functioning to the fact that their experience led them to re-examine their values and priorities in life, and to make sense of their experiences in a meaningful way. One father of a child with long term technology dependence described this well:

“It’s brought us closer in a lot of ways, too, because it gives you a different set of values, your career’s not as important, and things like that. It’s more emphasis on your family, and it changes your outlook on life a little bit, I think.” (O’Brien, 2001, pg. 17).

A father of a child with diabetes reflected on his role in the caregiving process as being important to maintaining a happy marriage and family as a whole:

“You have to be involved so the marriage is happier, the family is happier, because if it gets messed up, other things get messed up.” (Sullivan-Bolyai et al., 2006, pg. 29).

Examination of the various studies showed, on the whole, that families in which the father had an active caregiving or supportive role reported more positive influences on family functioning of caring for a child with chronic illness than families in which the mother was the sole or main caregiver. Interestingly, there only appeared to be two fathers who regarded themselves as the main caregiver across all of the studies in this review.

Internal Coping Strategies

The internal coping strategies parents used to manage their situation and associated emotions were reported in all of the studies. One of the coping strategies reported by parents in five of the studies (O’Brien, 2001; Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006; Rouf et al., 2012) was to adopt a state of ‘constant vigilance’ in order to always be prepared for any event that might occur. Although

some parents regarded it as a burden, it was regarded as a helpful strategy by many. This is depicted well by a father of a child with JRA:

“But I won’t let down my guard and think it’s gone away because it can always re-develop and re-flare up- you can never tell. It can be 10 years down the road; it could be 2 days, and you just have to be ready to deal with it.” (McNeill, 2004, pg. 533).

Acting as an advocate for their child was another coping strategy developed by many parents, especially in relation to hospital visits but also in other areas of life (O’Brien, 2001; Sallfors & Hallberg, 2003; Tong et al., 2010; Rouf et al., 2012). A parent of a child with CKD powerfully conveyed the necessity of acting as an advocate in the hospital, comparing it to being in battle:

“You go to the hospital and you’re a soldier for your child, you’re there mainly for him, you’re there for him, not for yourself...Sometimes they make it so difficult for you and it’s like I feel like you’re in a battle, and you’re battling for your child.” (Tong et al., 2010, pg. 551).

A mother of a child with a nut-allergy described having to continuously act as an advocate for her child and remind other parents and school of the importance of keeping the environment nut-free:

“I get a bit hot headed and I remind them (...) and I lay it, you know, I make it a big deal (...) so I’m the kind of, I’m the nut, you know, Hitler (laughs).” (Rebecca, 5:345). (Rouf et al., 2012, pg. 55).

Having hope and searching for positive meanings were other coping strategies employed by many parents across several studies (O’Brien, 2001; Sallfors & Hallberg, 2003; McNeill, 2004; Rouf et al., 2012). This is shown clearly in the following quotation by a mother of a child with JCA:

“I hope for a research breakthrough. I hope he’s going to be all right. There are lots of people who have lived with it for ages. Many of them have crippled joints, but I hope he won’t” (Sallfors & Hallberg, 2003, pg. 200).

Searching for positive meanings was particularly salient in fathers’ accounts. For example, a father of a girl with JRA discussed his outlook on his daughter’s illness, which appeared representative of the outlook of other fathers across several of the studies:

“I just think sometimes things happen for whatever reason they happen, and I believe [my daughter] to be very special. I believe she’s more special because she has a rare disease. And I think she’ll give back what she’s been given- tenfold.” (McNeill, 2004, pg. 536).

Other coping strategies mentioned less frequently in the studies included parents using downward comparison as a means of gaining perspective; living in the ‘here and now’ instead of thinking too much about the future; being emotionally detached when carrying out distressing medical procedures on their child; and becoming more actively involved in the medical care of their child in order to build confidence and empower themselves (Sallfors & Hallberg, 2003; Sullivan-Bolyai et al., 2006; Tong et al. 2010).

Co-Parenting

Parents in four of the studies (Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006; Tong et al., 2010) reported noticing a difference between how mothers and fathers coped with having a child with chronic illness, and a difference in the roles that mothers and fathers adopted as a result. In the study by McNeill (2004), most of the fathers of children with JRA reported feeling the need to be strong for the family, and assume the role of ‘protector’ in times of crisis. This led them to suppress their own needs and often not share their own feelings with their partner. Often, they did not have anyone else to turn to for support and relied excessively on self-support and internal coping strategies:

“Well, no, there wasn’t anyone that I could turn to other than my wife, and there was a sense that my wife needed more consoling than I did. So I suppressed my needs, feelings of fear and anxiousness about what my daughter was facing and the pain that she was going through.” (McNeill, 2004, pg. 534).

Additionally, fathers in several studies (Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006) felt that they maintained a more positive perspective than their partner and would often try to encourage their partner to adopt a more positive outlook:

“My wife and I have arguments, and there are times that my wife has negative thoughts, and I try to convince her to look at the positive side of it.” (McNeill, 2004, pg. 536).

Fathers’ perception across several studies was that this served to balance out their partner’s response and was a helpful support to their partner. Fathers in some studies also reported having more of a ‘just deal with it’ attitude that meant focusing on the things that they could change, and trying not to worry about the things out of their control (McNeill, 2004; Sullivan-Bolyai et al., 2006). This perception differed from maternal reports in other studies however, in which mothers reported a perception that fathers took a more passive role within the context of caring for their child’s chronic illness. Interestingly, according to some mothers, this appeared to be due to the mother’s need to be in control. For example, one mother of a child with JCA stated the following:

“I have taken the responsibility. My husband didn’t have a chance. I like it. I don’t let him in. It’s not that he hasn’t done his share, but if anything that I have robbed him of it. I know that’s what it’s all about. I think I can read her signals better than my husband, too” (Sallfors & Hallberg, 2003, pg. 197).

Fathers in this study reported perceiving their role to be more concerned with caring for the whole family, and often they would spend more time with healthy siblings and be the ‘providers’ for the family. This was similar to fathers of children with diabetes (Sullivan-Bolyai et al., 2006), who described the division of labour as ‘co-parenting’,

but reflected that the mothers carried out the main coordination and monitoring of their child's condition and care, with some fathers providing respite to the mothers in the evenings and weekends. Several fathers in this study reported that learning the care-skills from the start, and then staying involved in the child's care, was an important step in increasing their confidence and subsequently maintaining their involvement in caregiving:

"It's like nursing skills and if you don't do it you lose it. Fathers need to know that they need to be involved! It's hard for them to admit it and they are probably afraid to hurt the kid, where the moms are equally afraid, but someone has got to do it and if there was no mom I am sure they would" (Sullivan-Bolyai et al; 2006, pg. 29).

The reflection that fathers may not be as actively involved in caregiving as the mother due to a lack of confidence, or knowing that the mother will fulfil the caregiving role if they do not, was also a salient theme in the study by Tong and colleagues (2010). One mother in this study stated the following about the father:

"He doesn't want to see our daughter in pain, crying...he does not have to be in the room when I do the injections." (Tong et al., 2010, pg. 553).

Only a few mothers reported finding this difference in caregiving and coping frustrating; most mothers in the studies appeared to accept it as being a natural part of their family identity and functioning.

External Support

External support was raised as an important factor in parents' ability to cope in many studies, with parents reporting both negative and positive experiences with external support structures. Several parents across studies reported experiencing a lack of support from family, friends and other agencies due to a lack of understanding of their child's condition (O'Brien, 2001; Sallfors & Hallberg, 2003; Tong et al., 2010; Rouf et al., 2012). An illustration of this is provided by the mother of a child with a food allergy:

“Even if you tried to tell them they think you’re making it up (...) so they think nothing of trying to offer them sweets even though you’ve said to them can you not.” (Kimberley, 2: 454) (Rouf et al., 2012, pg. 58).

As a result, some parents described losing confidence in their support structures, and some reported feeling negatively evaluated by others due to the perception that they were lying about the severity of their child’s condition. Some parents reported initially not being able to use support from family and other agencies due to them not feeling confident in caring for the child. At the more extreme end, some parents reported that their family members did not even try to offer support, and would say hurtful things towards them and their child (O’Brien, 2001; Tong et al., 2010). One mother of a girl, who had become technology dependent following a near-drowning incident, relayed the following about her husband’s family’s reaction:

“And then his family is saying, ‘Put her away. Put her away. She’s a gork. She’s a vegetable. Put her away. What are you doing with her? What about your family?’ It was like, all of a sudden, this child who was so wonderful to her grandfather was dead.” (O’Brien, 2001, pg. 20).

Fortunately several parents also reported positive experiences of social support and stated that this was in large part what helped them cope with their situation. For example, one mother stated:

“We have families that are very supportive. That helps a lot, a lot. My parents and my husband’s parents both live like within 4 miles each. We wouldn’t have been able to do it without them.” (O’Brien, 2001, pg. 19).

Similarly, parents of children with JCA reported that being able to talk to others about their situation was very helpful:

“We have friends we can talk to. People who knew what he was like before he got sick. And we have the district nurse to talk to. Luckily.” (Sallfors & Hallberg, 2003, pg. 200).

Relationships with health care professionals were also mentioned as an important form of support in two studies (Sullivan-Bolyai et al., 2006; Tong et al., 2010). Parents reported feeling it was important for healthcare professionals to be accessible, open and consistent, which was not always their experience. Some parents indicated that it was important for healthcare staff to recognise their expertise in caring for their child, and that it was helpful when they used this to empower the parents (Sallfors & Hallberg, 2003; Tong et al., 2010). Additionally, many parents expressed that being able to see the same healthcare professional throughout their child's medical treatment was beneficial.

Helping the Child to Cope

The pain and psychological difficulties experienced by their child as a result of the chronic illness was a concern for several parents across studies. Parents across five of the studies discussed how important it was for them to help their child cope with their illness in order for them to feel 'normal' and enjoy the same activities their peers enjoyed (Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006; Tong et al., 2010; Rouf et al., 2012). Trying to keep the child's condition in perspective was central to this:

"To wrap (child) in cotton wool would (...) ruin his life and that's the last thing I want. He's a normal boy that can't eat nuts, he's not a walking nut allergy that happens to be a boy." (Elizabeth, 6: 186) (Rouf et al., 2012, pg. 58).

Parents in several studies reported using strategies such as normalising and becoming involved in activities so that their child could be involved too:

"We just got involved, constantly for her. We gave her so much support, she was able to participate in nearly everything. We kept things as normal as possible." (Sallfors & Hallberg, 2003, pg. 200).

Helping the child to be involved in activities with his/her peers and to feel normal often involved the parents hiding their own anxieties from their child so that the child did not think there was anything to worry about. This was described well by a father of a child with JRA:

"I don't want him to see that I am as worried about it as I am because I don't want him to be overly concerned about it. And if he's thinking about it all the time, then perhaps it's going to affect his everyday life. Maybe he's going to say "Maybe I shouldn't play baseball because I'm a little worried I'm going to hurt my knee, or maybe I don't want to do this in the gym"...he's going to think "if my father's really worried about it, there must be something to it."" (McNeill, 2004, pg. 537).

It appeared that focusing on how to help their child cope functioned as a coping strategy in itself for some parents. For example, a mother of a child with CKD described the following:

"You're sort of saving your strength just to help your child through it whereas if you start trying to face it, you just sort of fall apart and you're not strong enough for the child". (Tong et al., 2010, pg. 555).

Some parents reported that supporting their child to manage their condition could be challenging at times, especially when the parents were unable to differentiate between the child's illness behaviour and the child potentially using their illness as an excuse to escape certain duties or activities (Sallfors & Hallberg, 2003; Sullivan-Bolyai et al., 2006):

"It's difficult to know where the boundary line between ordinary teenage defiance and pain goes. Not forcing them to do things. I can look at her but I cannot feel her pain." (Sallfors & Hallberg, 2003, pg. 198).

Other parents were facing the challenge of how much to explain to their child about their condition and how soon, considering their child's developmental stage. The mother of a boy with a severe food allergy reflected on this:

"It's trying to get across the point of the severeness to him because we haven't gone into the fact that if he touches this he will die (...) he's got a child's eye view of dying (...) he's too young to have that conversation with and I don't really know, it would be

much older, but that will be quite hard explaining to him how serious it is” (Suzanne, 3: 468) (Rouf et al., 2012, pg. 57).

The child's developmental stage appeared to be a significant factor for parents across the studies, and clearly influenced the nature of parents' worries and coping strategies.

Discussion

Findings and Implications

A systematic search of the literature yielded 13 qualitative studies published between 2000 and 2014, exploring the parental experiences of caring for a child with chronic illness. They were rated for quality using the quality framework developed by Walsh and Downe (2006), leading to only six studies being of high enough quality to be included in the synthesis. Translating and synthesising the six studies led to the identification of six super-ordinate themes in the parental experience of caring for a child with chronic illness, which created a line of argument synthesis as described by Noblitt & Hare (1988): 1) Emotions 2) Impact on Family Functioning 3) Internal Coping Strategies 4) Co-Parenting 5) External Support and 6) Helping the Child to Cope.

The main emotions identified by parents directly following diagnosis involved feelings of shock, devastation, loss, grief, despair, anxiety, fear and sadness. This is similar to findings in earlier research (e.g. Brown et al., 2008; Coffey, 2006) and is understandable given the context in which they are experienced. Interestingly, some of the emotions reported in Coffey's (2006) meta-ethnography as being reported frequently by parents following diagnosis, specifically severe depression and suicidality, were hardly mentioned by parents in the current studies. It could be speculated that this may reflect the increasing awareness of paediatric chronic illness amongst parents and healthcare providers, and the associated increased support available when parents are told about their child's diagnosis.

The most common emotions reported as being experienced daily by parents included anxiety, frustration, powerlessness, hope, despair, and sadness. Again, this is similar to what has been demonstrated previously in the literature (Cousino &

Hazen, 2013; Brown et al., 2008). The studies in this synthesis emphasised how important it is for parents to feel that they have some control over their child's illness, and that managing their feelings of helplessness and uncertainty is one of the most difficult parts of their experience. This is important for healthcare professionals to consider, and indicates that healthcare professionals could have a role to play in terms of empowering the parents and helping them to feel more in control of their child's condition where possible.

The impact on family functioning of caring for a child with chronic illness appeared to be significant in most parents' experiences. Parents' reports of the way in which their child's chronic illness had impacted their family, however, varied across studies. The negative impact of childhood chronic illness on family functioning is well documented in the literature (Brown et al., 2008; Cousino & Hazen, 2013; Coffey, 2006), and was mainly reported by parents of children with CKD and parents of children with long-term technology dependence in the current review (O'Brien, 2001; Tong et al., 2010). The positive impact on family functioning, as perceived by many parents in the current studies, has been less well documented and, to the author's knowledge, has not been discussed in previous literature reviews on the topic. This is a new and interesting insight into the parental experience of caring for a child with chronic illness- namely that parents feel that their child's chronic illness has brought the family closer together, has led them to re-adjust their values in life, and has given them a different perspective on what is important. Being able to view their experience in this way can be considered a protective factor, and one that will most likely foster resilience in the parents and their family. Examination of the various studies showed, on the whole, that families in which the father had an active caregiving or supportive role reported more positive influences on family functioning than families in which the mother was the sole or main caregiver. This makes sense considering the vast array of literature that shows how important social support is in helping individuals to cope with stress (e.g. Thoits, 1995).

The synthesis revealed that parents reported similar internal coping strategies across studies, specifically maintaining a state of constant vigilance and acting as an advocate for their child. These have also been reported as coping strategies in previous studies (Coffey, 2006; Cohen, 1995; Ray & Ritchie, 1993; Austin, 1991).

The finding that many parents still needed to be assertive and act as an advocate regarding their child's needs within the hospital and school environment was concerning, given that this has been noted many times in the earlier literature and indicates a clear need for service improvement. The current review found that parents' lack of control, combined with poor communication, insufficient information and lack of time with healthcare professionals resulted in increased uncertainty and anxiety (Sallfors & Hallberg, 2003). According to the data in the studies, the most appreciated professionals are those who take time to listen to the parents and who explain things to them in a way they can understand (Sallfors & Hallberg, 2003). Additionally, the studies found that parents would like to experience a collaborative, equal partnership with clinicians, and expect clinicians to respect their expertise on their child and his/her condition (O'Brien, 2001; Sallfors & Hallberg, 2003; Tong et al., 2010).

Maintaining hope and searching for positive meanings were additional coping strategies reported by parents in the studies. Hope has previously been found to correlate positively with adjustment in mothers of children with chronic illness (Wallander & Varni, 1998). Interestingly, searching for positive meanings was particularly salient in fathers' accounts. McNeill (2004) discusses that the group of fathers he interviewed seemed to view seeking a silver lining as part of their role. Previous research has found that mothers experience poorer mental health than fathers in families with a child with chronic illness (Manuel, 2001; Thompson & Gustafson, 1996). Therefore, McNeill's (2004) discovery is significant given that the findings on meaning of illness consistently show that having a more positive cognitive representation of illness is associated with more effective adaptation (Goode, Haley, Roth et al., 1998; Heru, 2000). Additionally, earlier research has found that mothers' perceptions of child illness are influenced by fathers' perceptions (Frey, Fewell & Vadasy, 1989), implying that in this case, mothers' perceptions may become more positive over time, due to the influence of the fathers.

Fathers in one of the studies (Sullivan-Bolyai et al., 2006) described the division of responsibilities between mother and father as 'co-parenting', but reflected that the mothers carried out the main practical and emotional aspects of their child's care. This seemed consistent with paternal and maternal reports in several of the other

studies (Sallfors & Hallberg, 2003; McNeill, 2004; Tong et al., 2010). Additionally the studies in the synthesis indicated that fathers may be more emotionally attuned than has previously been considered (McNeill, 2004), but choose to hide their emotions from their partner and child in order to help them cope and to act as the 'protector'; a role that the fathers seem to adopt naturally.

Interestingly, some mothers reported a perception that fathers took a more passive role within the context of caring for their child's chronic illness due to the mothers' need to be in control (Sallfors & Hallberg, 2003). Related to this, several fathers in one study reported that learning caregiving skills from the point of diagnosis, and then staying involved in the child's care, was an important step in increasing their confidence and subsequently maintaining their involvement in caregiving (Sullivan-Bolyai et al., 2006). This indicates that fathers may initially lack confidence in their caregiving skills and find this difficult to communicate, leading to the mothers taking control over the caregiving responsibilities and somewhat excluding the fathers. This insight emphasises the importance of healthcare staff actively involving the father in the child's care from the start, and making efforts to include fathers who may present as less willing or lacking confidence in their role (Sallfors & Hallberg, 2003).

McNeill (2004) suggests that understanding the various coping strategies used by mothers and fathers is more helpful if examined in the context of the couple relationship rather than from an individual perspective, and the current synthesis would support this supposition. When examined through a relational lens, it appears that mothers' and fathers' different coping strategies often can and do complement each other and, as demonstrated by some of the studies in this synthesis, foster resilience in the family as a whole (Sallfors & Hallberg, 2003; McNeill, 2004; Sullivan-Bolyai et al., 2006).

Positive social support has consistently been shown to buffer the effects of stress (Thoits, 1995). This synthesis disclosed that many parents have experienced negative as well as positive encounters with external sources of support. For example, several parents reported that other people and agencies did not have an understanding of their child's condition and/ or lacked confidence in their ability to care for the child, making it difficult for the parents to trust others or leave the child in

their care. These experiences indicate that it may be important for healthcare staff to ensure improved communication with other agencies and support structures, in order to provide information on the child's condition and how to support the child and family. Ensuring someone adopts a 'liaison' role within relevant healthcare teams may help to foster this type of improved communication in the future.

The synthesis revealed the challenge faced by parents in supporting their child to manage their condition, especially when trying to differentiate between the child's illness behaviour and the child potentially using their illness as an excuse to escape certain duties or activities (Sallfors & Hallberg, 2003; Sullivan-Bolyai et al., 2006). Consideration of the child's developmental stage was also a significant factor for many parents: firstly in deciding when to give the child more responsibility regarding the management of their condition, and secondly in considering developmental issues of adolescence such as relationships, independence and future vocation (O'Brien, 2001). Healthcare professionals could have a role here in providing information and support to the parents regarding the child's developmental stage and subsequent level of expected understanding/ independence. Preparing parents for key points and transitions in the child's development may also be a helpful way to support parents with this challenge (O'Brien, 2001; Tong et al., 2010).

The findings from this synthesis suggest several pointers for service improvement. It is recognised that parents' perspectives of an encounter or of service delivery may differ from clinicians' perspectives of the same encounter or service, and that some of the suggestions made here may be difficult to achieve when operating within a tight budget. Nevertheless, there is a growing consensus that the needs, preferences and experiences of service users should be considered in the development and evaluation of service delivery models (Ring et al., 2010) and this synthesis helps to further our understanding of parents' views of the services they use in relation to their child with chronic illness.

Limitations

A great majority of the participants in the current studies appeared to have a similar demographic profile: most parents were in full or part-time employment, white, well-

educated, relatively affluent, and English speaking. McNeill (2004) explicitly states in his study:

“Of the fathers who were invited to participate in the study but refused, it is worth noting that the majority were either visible minorities or had lower socioeconomic status. Reasons given were that they were not interested in participating or were too busy”. (pg. 530).

Additionally, most of the fathers who participated in the research studies viewed themselves as having an active caregiving role, and this may differ from the roles of fathers who did not participate. Thus, it is important to note that the data gathered from these studies may only reflect the experiences of parents with a specific demographic profile and/ or parenting role.

References

- Austin, J.K. (1991). Family adaptation to a child's chronic illness. *Annual Review of Nursing Research*, 9, 103-120.
- Brown, R., Wiener, L. Kupst, M., Brennan, T., Behrman, R., Compas, B., Elkin, D., Fairclough, D., Friebert, S., Katz, E., Kazak, A., Madan-Swain, A., Mansfield, N., Mullins, L., Noll, R., Patenaude, F., Phipps, S., Sahler, O., Sourkes, B. & Zeltzer, L. (2008). Single Parenting and Children with Chronic Illness: An Understudied Phenomenon. *Journal of Pediatric Psychology*, 33(4), 408–421.
- Case-Smith, J. (2004). Parenting a child with a chronic medical condition. *American Journal of Occupational Therapy*, 58, 551-560.
- Centre for Research and Dissemination (2008). *Systematic Reviews. CRD's guidance for undertaking reviews in healthcare*. York: York Publishing Services Limited
- Coffey, J.S. (2006). Parenting a child with chronic illness: A metasynthesis. *Journal of Pediatric Nursing*, 32 (1), 51- 59.
- Cohen, M.H. (1995). The triggers of heightened parental uncertainty in chronic, life-threatening childhood illness. *Qualitative Health Research*, 5, 63-77.
- Cousino, M. & Hazen, R. (2013). Parenting Stress among Caregivers of Children with Chronic Illness: A Systematic Review. *Journal of Pediatric Psychology*, 1–20.
- Dixon-Woods, M., Shaw, R.L., Agarwal, S. & Smith, J.A (2004). The problem of appraising qualitative research. *Quality and Safety in Health Care*, 13, 223-225.
- Frey, K., Fewell, R., & Vadasy, P. (1989). Parental adjustment and changes in child outcome among families of young handicapped children. *Topics in Early Childhood Special Education*, 8 (4), 38-57.
- Gannoni, A.F. & Shute, R.H. (2010). Parental and child perspectives on adaptation to childhood chronic illness: A qualitative study. *Clinical Child Psychology and Psychiatry*, 15 (39), 39-53.
- Goode, K., Haley, W., Roth., D. & Ford, G. (1998). Predicting longitudinal changes in caregiver physical and mental health: a stress process model. *Health Psychology*, 17(2), 190-198.
- Heru, A. (2000). Family functioning, burden and reward in the caregiving for chronic mental illness. *Families, Systems & Health*, 18 (1), 91-103.
- Maciver, D., Jobes, D. & Nicol, M. (2010). Parents' experiences of caring for a child with chronic pain. *Qualitative Health Research*, 20 (9), 1272-1282.
- Maltby, H.J., Kristjanson, L., Coleman, M.E. (2003). The parenting competency framework: Learning to be a parent of a child with asthma. *International Journal of Nursing Practice*, 9, 368-373.

Manuel, J. (2001). Risk and resistance factors in the adaptation in mothers of children with juvenile chronic arthritis. *Journal of Pediatric Psychology*, 26 (4), 237-246.

McNeill, T. (2004). Fathers' experience of parenting a child with juvenile rheumatoid arthritis. *Qualitative Health Research*, 14 (4), 526-545.

Mokkink, L.B., van der Lee, J.H., Grootenhuis, M.A, Offringa, M., & Heymans, H.S.& The Dutch National Consensus Committee "Chronic Diseases and Health Conditions in Childhood" (2008). Defining chronic diseases and health conditions in childhood (0–18 years of age): national consensus in the Netherlands. *European Journal of Pediatrics*, 167, 1441–1447.

Noblitt, G.W & Hare, R.D. (1988) *Meta-Ethnography: Synthesizing Qualitative Studies (Qualitative Research Methods Series II)*. London: Sage Publications Ltd.

O'Brien, M. (2001) Living in a house of cards: family experiences with long-term childhood technology dependence. *Journal of Pediatric Nursing*, 16 (1), 13-22.

Pitchforth, E., Weaver, S., Willars, J., Wawrzkowicz, E., Luyt, D. & Dixon-Woods, M. (2011). A qualitative study of families of a child with a nut allergy. *Chronic Illness*, 7(4), 255-266.

Ray, L.D. & Ritchie, J.A. (1993). Caring for chronically ill children at home: Factors that influence parents' coping. *Journal of Pediatric Nursing*, 8, 217-225.

Ring, N., Ritchie, K., Mandava, L. & Jepson, R. (2010). A guide to synthesising qualitative research for researchers undertaking health technology assessments and systematic reviews. Available from: <http://www.nhshealthquality.org/nhsqis/8837.html> [Accessed online in July 2013]

Rouf, K., White, L., & Evans, K. (2012). A qualitative investigation into the maternal experience of having a young child with severe food allergy. *Clinical Child Psychology & Psychiatry*, 17 (1), 49-64.

Sallfors, C., & Hallberg, L. R.. (2003). A parental perspective on living with a chronically ill child: A qualitative study. *Families, Systems, & Health*, 21 (2), 193-204.

Spencer, L., Ritchie, J., Lewis, J. & Dillon, L. (2003). *Quality in Qualitative Evaluation: A framework for assessing research evidence*. Government Chief Social Researcher's office: Crown Copyright.

Sullivan-Bolyai, S., Deatrick, J., Gruppuso, P., Tamborlane, W. & Grey, M. (2003). Constant vigilance: Mothers' work parenting young children with type 1 diabetes. *Journal of Pediatric Nursing*, 18 (1), 21- 29.

Sullivan-Bolyai, S., Rosenberg, R., & Bayard, M. (2006). Fathers' reflections on parenting young children with type 1 diabetes. *American Journal of Maternal Child Nursing*, 31 (1), 24-31.

Thoits, P.A. (1995). Stress, coping and social support processes: where are we? What next? *Journal of Health and Social Behaviour, Extra Issue*, 53-79.

Thompson, R.J. & Gustafson, K.E. (1996). *Adaptation to chronic childhood illness*. Washington, DC: American Psychological Association.

Todd, M., Welsh, J. & Moriarty, D. (2002). The experience of parents of children with primary lymphoedema. *International Journal of Palliative Nursing*, 8 (9), 444-451.

Tong, A., Lowe, A., Sainsbury, P., & Craig, J. C. (2010). Parental perspectives on caring for a child with chronic kidney disease: An in-depth interview study. *Child: Care, Health & Development*, 36 (4), 549-557.

Van der Lee, J.H., Mokkink, L.B., Grootenhuis, M.A., Heymans, H.S. & Offringa, M. (2007). Definitions and Measurement of Chronic Health Conditions in Childhood: A Systematic Review. *Journal of the American Medical Association*, 297 (24), 2741-2751.

Wallander, J.L. & Varni, J.W. (1998) Effects of pediatric chronic physical disorders on child and family adjustment, *Journal of Child Psychology and Psychiatry*, 39, 29-46.

Walsh, D. & Downe, S. (2006). Appraising the quality of qualitative research. *Midwifery*, 22, 108-119.

Chapter 2: Major Research Project

Caregivers' Experiences of Caring for a Child with Cardiac Arrhythmia who has an Automatic External Defibrillator: An Exploratory Study using Interpretative Phenomenological Analysis

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Prepared in accordance with guidelines for submission to the Journal of Pediatric Psychology (Appendix 5)

PLAIN ENGLISH SUMMARY

BACKGROUND

'Cardiac arrhythmia' describes a condition where the heart muscles contract either too slowly, too quickly, or irregularly. There are some children with cardiac arrhythmia who are at risk of their heart suddenly stopping. The hospital gives families of these children a defibrillator and teaches them how to use it in the event of their child's heart stopping, to decrease the risk of sudden death. A defibrillator is an automated machine which can re-start the child's heart using an electrical shock if it is placed on the child's chest.

AIM

The aim of this study was to explore the experiences of caregivers caring for a child with cardiac arrhythmia who has a defibrillator.

METHODS

Main caregivers of children with cardiac arrhythmia who had a defibrillator were asked to take part in the study. Caregivers were identified by their child's cardiologist at RHSC, Yorkhill Hospital, and all eligible caregivers were sent a letter asking them to take part in the research. There were only 16 families in Scotland who had a child with cardiac arrhythmia and who had a defibrillator. Out of these 16 families, seven main caregivers (two were couples) agreed to take part in the research. The researcher carried out in-depth interviews with them, lasting about one hour each. A qualitative research design was used to gain an in-depth understanding of the caregivers' experiences. Interviews were digitally recorded, typed up and then analysed by the researcher to explore shared experiences and differences in the experiences of the caregivers. These were then presented in the form of a written report. All information from the interviews was kept anonymous.

MAIN FINDINGS AND CONCLUSIONS

The research identified several common themes in the experiences of the caregivers. These related to the impact of the cardiac arrhythmia on the families' lives, the impact of having a defibrillator on the families' lives, their experiences with hospital support, and the coping strategies they used to manage their daily experiences. It

became apparent that caring for a child with cardiac arrhythmia can lead to feelings of distress and worry for caregivers, especially because there is a large element of uncertainty with the condition. Being given a defibrillator increased the anxiety levels of some caregivers; however others felt relief at being given one. Some caregivers had difficulty ensuring their child's school obtained a defibrillator, which was concerning. Overall, caregivers had very positive experiences with hospital support however there were some difficult incidents reported related to poor communication from hospital staff.

The findings from this study will be used to inform services about how they can best support children with cardiac arrhythmia who have a defibrillator, and their families. A Clinical Psychology service within the RHSC cardiology department is currently being developed, and this research will help directly inform the new service.

ABSTRACT

Objective: The prevalence of paediatric cardiac arrhythmias is increasing, and management of some high-risk arrhythmias now involves giving the family an automatic external defibrillator (AED) to use in the case of their child experiencing sudden cardiac arrest. No earlier research has explored caregivers' experiences of caring for a child with cardiac arrhythmia who has an AED. This qualitative study aimed to explore the experiences of caregivers caring for a child with cardiac arrhythmia who has an AED.

Methods: In-depth semi-structured interviews were conducted with seven caregivers (two couples) across five interviews. Interpretative Phenomenological Analysis was used to analyse the data.

Results: Four super-ordinate themes were identified and, due to word limit restrictions, three have been reported on here: the impact of cardiac arrhythmia on daily life, experiences of living with the AED, and experiences of hospital support.

Conclusions: A greater understanding of caregivers' experiences in this context will be used to inform services about how they can best support and meet the needs of children with cardiac arrhythmia who have an AED, and their families.

Keywords: Paediatric cardiac arrhythmia; caregiver experiences; Paediatric chronic illness; interpretative phenomenological analysis; qualitative research.

INTRODUCTION

Cardiac Arrhythmia

Over 700,000 people in the UK have a cardiac arrhythmia (Department of Health, 2005). Cardiac arrhythmia is an umbrella term used to describe a number of conditions where the heart muscle contracts either too slowly (called bradycardia), too quickly (called tachycardia), or irregularly because of a disturbance in the heart's normal electrical activity (Department of Health, 2005). This can be due to a genetic disorder or acquired condition (Hanash & Crosson, 2010). Consequences of arrhythmias can range in severity from minor discomfort to the risk of sudden death (Department of Health, 2005). Although many arrhythmias are benign, they may significantly affect a person's quality of life. Symptoms include fainting, light headedness, dizziness, sensations of heart flutters, shortness of breath, chest-pain, weakness, fatigue, and intolerance for activity (American Heart Association, 2013).

The exact prevalence of paediatric arrhythmias is unknown. They are less common than in adults, but are increasing due to successful repair of congenital heart disease, which can leave the surviving child with an arrhythmia (Doniger & Sharieff, 2006). Types of arrhythmias found in children include Supraventricular tachycardia, sinus bradycardia, Long Q-T Syndrome (LQTS), Wolff-Parkinson-White Syndrome, Ventricular Tachycardia, Sick Sinus Syndrome and Complete Heart Block (American Heart Association, 2013). Treatment of these arrhythmias varies widely according to cause and severity, and can include lifestyle changes, medication, use of a pacemaker, external cardiac defibrillation, an implanted cardioverter defibrillator (ICD), cardiac ablation, and surgery (American Heart Association, 2013).

Cardiac Defibrillation

Cardiac defibrillation is essential if the child experiences a cardiac arrest. Survival rates decrease significantly with the passage of time from cardiac arrest to defibrillation (Samson et al., 2003). For this reason, children who are deemed to be at highest risk of sudden death are fitted with an ICD. There is an additional population of children with cardiac arrhythmia who are also deemed to be at increased risk of sudden death, however not high enough risk to warrant an ICD due to the risks associated with implanting and maintaining such technology (Divekar & Soni, 2006). For these children, families are offered and trained to use an automatic

external defibrillator (AED). The practice of paediatric external defibrillation is mainly derived from adult studies, and this is an on-going area of development in paediatric care. Questions therefore remain regarding optimal techniques for paediatric defibrillation and management (Haskell & Atkins, 2010).

A significant body of quantitative research already exists in relation to the experience of living with cardiac arrhythmia and an ICD (e.g. De Maso et al., 2004; Dunbar et al., 2012). Additionally, a qualitative study carried out by Rahman and colleagues (2011) examined the experience of parents caring for a child with an ICD, and their children's experience of living with the ICD. Parents of children with cardiac arrhythmia and an ICD, however, will arguably have a different experience from parents of children with cardiac arrhythmia and an AED. A qualitative study carried out in the USA (Farnsworth et al., 2006) explored parental perceptions concerning congenital LQTS, a form of cardiac arrhythmia. Findings indicated that parents with young children were afraid of their child dying, and some found that carrying an AED helped to alleviate their fear; however, the study did not explore this aspect of their experience further. Surprisingly, no other qualitative literature appears to exist specifically exploring the experience of parents caring for a child with cardiac arrhythmia and an AED.

There is some research examining the effect on adult patients and their family members of being provided with an AED and this shows mixed results. One small study involving interviews with post myocardial infarction (MI) patients found that AEDs were highly valued by patients and their partners, and increased perceived control over the heart disease (Chen, Eisenberg & Meischke, 2002). Slightly different results were found in a randomised study by Cagle et al. (2007) comparing quality of life in patients assigned to either cardiopulmonary resuscitation (CPR) or CPR/AED training. They found that the CPR/AED group reported worse scores on quality of life, particularly in those subscales relating to social functioning. Finally, a larger longitudinal observational study, comparing the long-term effects of CPR training and CPR/ AED training on anxiety and depression of patients and their partners, found that anxiety of partners in the CPR/AED group increased slightly over time, whereas for the CPR group, partners' anxiety decreased significantly over the 2 years of follow-up (Thomas et al., 2011). The authors consequently recommended the

assessment of anxiety in partners of patients who receive AEDs, and consideration of strategies to reduce their anxiety (Thomas et al., 2011). In contrast to the study by Cagle et al. (2007), Thomas et al. (2011) found no evidence to suggest that home AEDs caused distress among the patients. Despite mixed results, these studies suggest that a patient's and caregiver's experience of having an AED is worthy of further exploration.

Considering the above findings and also a caregiver's role in caring for a child with a chronic condition such as cardiac arrhythmia, the importance of exploring caregivers' experiences of caring for a child with cardiac arrhythmia who has an AED becomes apparent. A comprehensive review of qualitative research exploring the experiences of parents caring for a child with chronic illness can be found in the meta-ethnography by Anker-Petersen (2014). In general, the literature suggests a reciprocal relationship between chronic illness and parental adaptation, whereby the child's illness impacts on parents' functioning and parental functioning impacts on child adaptation (Brown et al., 2008).

As cardiac arrhythmia arguably differs from other chronic illnesses, and the experience of having a child with an ICD will differ from having a child with an AED, it is important to gain an understanding of caregivers' experiences of caring for a child with cardiac arrhythmia who has an AED. No literature exists, to the author's knowledge, exploring the experiences of caregivers caring for a child with cardiac arrhythmia who has an AED. A greater understanding of caregivers' experiences in this context can be used to inform services of how best to support and meet the needs of children with cardiac arrhythmia who have an AED, and their families. As suggested in the literature, caregiver coping may impact upon child coping, and this is another key reason for exploring caregivers' experiences within this population.

AIM

This study aimed to explore the experiences of main caregivers caring for a child with cardiac arrhythmia who has an AED.

METHOD

Ethical Considerations

The study was approved by the University of Glasgow, Greater Glasgow and Clyde Research and Development Team, and the NRES Committee East Midlands (see appendices 6-8). Confidentiality was carefully discussed with participants and written consent obtained prior to the interviews. Participants were informed that they had the right to withdraw at any time, with no impact on their child's medical treatment. All participants were informed that a clinical psychologist was available for support should they experience any distress during or after the interview; however this was not required for any participants.

As data collection progressed it became apparent that it would be difficult to ensure complete anonymity for some participants if the context and rich detail of their accounts was to be truly represented in the final report (see Kaiser, 2009, for a discussion on the dilemmas of protecting confidentiality in qualitative research). The researcher therefore contacted participants via telephone to discuss this issue with them, or discussed it at interview. The participants have given their explicit consent regarding the details included in this final report.

Design

This study is qualitative and used Interpretative Phenomenological Analysis (IPA). IPA aims to explore an important aspect of the research participant's life, for example their experience of an illness, from their personal perspective, whilst recognising that how this account is interpreted by the researcher(s) is inevitably influenced by their own cognitions and past experiences, plus the process of interaction between the researcher(s) and the participant (Smith & Eatough, 2007).

IPA is rooted in the theories of phenomenology, hermeneutics and idiography. To elaborate, phenomenology is concerned with understanding lived experience, which is a central focus of IPA. Along with this, IPA recognises that research inevitably involves a process of interpretation from the researcher. This is the way in which IPA draws on the hermeneutic approach (Smith, 2011). Smith (2011) describes the process of IPA as “engaging in a *double hermeneutic*, whereby the researcher is trying to make sense of the participant trying to make sense of what is happening to them” (pg. 10, italics taken from original). Finally, IPA has an idiographic commitment to analyse each participant's reports of their lived experience in-depth.

When several participants' accounts are drawn upon, IPA involves a detailed examination of each case followed by the search for patterns across cases (Smith, 2011). For a more comprehensive overview on the theoretical underpinnings of IPA, please see the paper by Shinebourne (2011).

Due to the focus of IPA being an in-depth idiographic examination of lived experience, it was felt that IPA was the most appropriate qualitative method to use for this study. Smith, Flowers, and Larkin (2009) provide an overview of the similarities and differences between IPA and other qualitative methodologies if the reader wishes to examine this further.

Sample

In accordance with the requirements of IPA, a purposive and well defined sample of parents for whom the research question was meaningful was used (Smith & Eatough, 2007). At the time of undertaking the research, there were only 16 families in Scotland who had a child or children with cardiac arrhythmia, with an AED. It was hoped to recruit between 6 and 8 main caregivers from this population in order to maintain an idiographic approach in line with IPA's philosophy. Small sample sizes are recommended to allow for detailed case by case analysis, to form an in-depth and rich understanding of the perceptions and experiences of the participants involved (Smith et al., 2009).

The main caregivers from all 16 families were invited to take part in the research, and six families responded. Of the respondents, one caregiver failed to attend the interview and subsequently was not contactable. Therefore, the final sample consisted of five families. Two families identified both parents as main caregivers and wished to be interviewed together. As a result, seven main caregivers were interviewed across five interviews. This number of interviews is consonant with the advice given to those undertaking professional doctorates (Smith et al., 2009 pg.52) Please see Table 1 for an overview of participant demographics.

Table 1. Participant Demographics

Interview No	Pseudonyms	Relationship to child	Relationship Status	Age of child now (years)	Diagnosis of child	Age at diagnosis (years)	Age when AED received (years)
1	Jane and Jack	Mother and Father	Married	17	Long QT Syndrome	6	6
2	Lisa and Grant	Mother and Father	Married	6	Cardiac Arrhythmia with identified gene mutation	Pre-birth	4
3	Terri	Grandmother	Single	12, 15 & 17	Long QT Syndrome	2, 5 & 7	5, 8 & 11
4	Mary	Mother	Married	15 & 19	Long QT Syndrome	11 & 11	9 & 13
5	Jennifer	Mother	Married	5	Cardiac Arrhythmia	5 weeks	5 weeks

Recruitment Procedures

Eligible participants were identified by the cardiac team at Yorkhill Hospital. The consultant cardiologist sent eligible participants a study information letter from the researcher. Caregivers who were interested in participating completed and returned a participant response form with their contact details. The researcher subsequently contacted them to answer any questions they had regarding the research. Following this, appointments were arranged to conduct the interview in a hospital out-patient department. One participant lived on a remote Scottish Island and therefore could not attend the interview in person; therefore a telephone interview was arranged. See appendices 9-12 for forms used during recruitment.

Research Procedures

Open-ended semi-structured interviews of approximately one hour were conducted with each family, with interviews lasting between 49 and 85 minutes. An interview schedule informed by the existing literature was devised to guide the interviews (see appendix 13). In accordance with guidance (Smith et al., 2009, pg.64) the researcher used the schedule flexibly, allowing exploration of other areas raised by the participants which were not on the schedule. Interviews were recorded on a

digital voice recorder and then transcribed verbatim by the researcher, with all identifiers of person and place removed.

Data Analysis

Data analysis was conducted in accordance with the IPA steps outlined by Smith et al (2009); please see Table 2. Two transcripts were independently analysed by both the researcher and the project supervisors, and themes resulting from the independent analyses were compared to check the validity of the primary analysis. Agreement was achieved in terms of the content of salient themes.

Table 2. Steps of IPA analysis, compiled from text in Smith et al. (2009, pages 79-101)

Step	Description of process
1: Reading and re-reading	Reading and re-reading of the transcript to immerse oneself in the data and become actively engaged with the participants' world.
2: Initial noting	Examination of semantic content and language use leading to initial notes of anything of interest within the transcript.
3: Developing emergent themes	Develop emergent themes within the transcript by mapping the relationships and patterns across and between exploratory notes.
4: Searching for connections across emergent themes	Develop a mapping of how the researcher thinks the themes fit together to produce a structure which outlines the most interesting and important aspects of the participant's account.
5: Moving to the next case	Repeat steps 1-4 for the next transcript, treating the next case on its own terms.
6: Looking for patterns across cases	Look for patterns and connections across cases to produce super-ordinate themes and emergent themes which represent all of the data.

Researcher Reflexivity

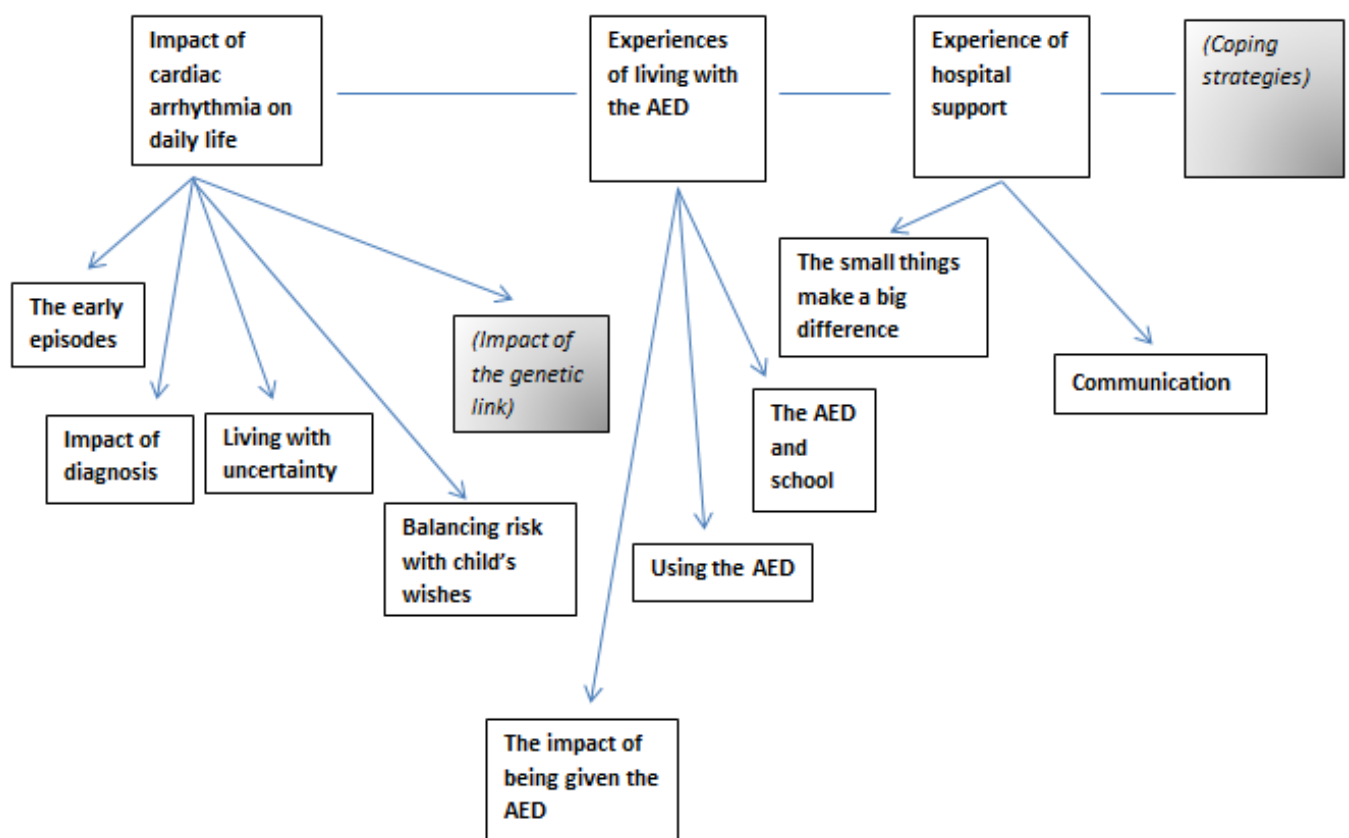
In IPA there is acknowledgement that the researcher brings their own beliefs and experiences into their interpretation of the data (Smith & Eatough, 2007). Therefore, the researcher spent time considering their own beliefs and experiences and how these might influence their interpretation, in addition to how they might influence the participants' ability to engage in the interview process. The researcher had previously worked clinically with parents of children with various chronic illnesses, and therefore had developed a level of clinical insight into the typical challenges faced by the paediatric population. This may have facilitated engagement and rapport with participants. The researcher had not worked with parents of children

with cardiac arrhythmia before and therefore was coming to this particular area of illness experience with little prior clinical knowledge or preconceptions. The researcher was aware that the research could potentially have an impact on them emotionally and therefore kept a personal reflective diary throughout the research process. They additionally used supervision to reflect on the process and emotional impact of the interviews in order to assess the influence of this on their interpretations of the data, and to contextualise their findings.

RESULTS

This study focused on exploring the experiences of caregivers caring for a child with cardiac arrhythmia who has an AED. A number of themes were identified as outlined in Figure 1.

Figure 1. Super-ordinate and emergent themes



The themes in the non-shaded boxes will each be explored; due to word count limitations, however, the bracketed themes in the shaded boxes will not be discussed. This is in line with guidance by Smith (2011) who recommends that sufficient space must be given to the elaboration of each theme, and therefore it may be better to present a subset of themes in some cases. Some of the coping strategies employed by the caregivers will be discussed due to their close link with other emergent themes; however space did not allow full discussion of all coping strategies used. Findings related to the bracketed themes have been well documented in earlier published studies (*cf.* Anker-Petersen, 2014; Andersen et al., 2008) and therefore it seemed appropriate to omit these themes as they add nothing to the knowledge base. Appendix 14 provides an overview of the omitted themes.

Impact of cardiac arrhythmia on daily life

Cardiac arrhythmia affected daily life for all the families in different ways. The following themes related to this will be explored: the early episodes; the impact of diagnosis; living with uncertainty; and balancing risk with their child's wishes.

The Early Episodes

The families described different pathways to diagnosis: children were diagnosed at varying ages and some experienced several undiagnosed seizures pre-diagnosis. One experience that appeared salient for three of the families was the effect of early episodes or seizures on the caregivers' emotions and adjustment. One father, whose son had nine seizures prior to being diagnosed at age six, described the trauma of experiencing these:

"I think you know you talked about what it's like for parents, and particularly those episodes, those early episodes, were just terrifying, you know? To, to not know what this was, that was causing your child to be, you know, collapsing, screaming out, collapsing, and then having this kind of fit [] Just hoping, *praying* that he would come round, that he would respond you know? Cause the eyes were rolled in the back of his head, and (deep sigh)...[] I think it's, on reflection, realising how close you'd come to losing him. [] Em, became eh, became just something that we were really *really* aware of, you know..." (Jack)

Jack powerfully conveys how traumatic it felt for him and his wife to see their son experience a seizure. His description of not knowing what 'this' was infers that he regarded the cause almost as an external force that they had no control over, emphasising their powerlessness in the situation. His vivid recollection of his son's appearance and behaviour during these seizures accentuates the traumatic nature of them. Jack movingly portrays their desperation and hope during each seizure that their son would "come round" again. Being aware of how close they had come to losing their son was a poignant recurring theme for Jack and Jane, and was also a theme echoed throughout the interview with Jennifer, whose son experienced a cardiac arrest at 5 weeks old:

"Oh it was traumatic, em awful. You know we didn't know what was wrong with him. And eh, we got, well we live up on [Anon Island] so we were in [Anon hospital] and then he was flown away. And eh, probably the worst thing at the time was we couldn't go with him because of eh you know, there was sort of 4 or 5 staff with him and there wasn't room for us and had something else happened on route..." (Jennifer)

For Jennifer, the circumstances surrounding her son's cardiac arrest added to the trauma. Also, not knowing what was wrong with her son made the whole experience more difficult, a similar reflection to that made by Jack.

Lisa and Grant were aware of their son's condition prior to his birth, and so were aware of the cause of his seizures from the start. Reflecting on their son's early seizures, Lisa described the following:

"Yeah, well it has, it has kinda had knock on effects for us at times. I mean certainly in the early years when he was not well it sort of kinda [] it affects you in a way that you don't think it's affecting you. But, I think when you look back on it, every time he's had a major upset and he's had to be put in hospital, it takes a bit off you. You know, it does, it wears you out a little bit. And then, cause I think as soon as he's fine, you go home, you're just so kinda like 'oh right I'm home!' and everybody's going back to normal, but, you know, [] it *totally* upsets *everybody*." (Lisa)

Lisa's description shows her struggle to realise how much the early seizures had affected her and her family at the time, and it is only on reflection that she has

become aware of the significant impact they had on everyone. It appears she might have been unable to process the impact of their experience at the time due to the way in which life continued as 'normal' as soon as their son had recovered and returned from hospital. This begs the question of how much impact a modern lifestyle and its accompanying demands might have on influencing a family's ability to process and adjust to a child's long-term illness. Lisa's description conveys emotional exhaustion and indicates a decrease in her ability to cope with the situation, emphasising the accumulative impact of her son's seizures.

The Impact of Diagnosis

Receiving the diagnosis itself was a significant turning point for four of the families, and had both short and long-term consequences for various family members. Terri, who had not suspected there to be anything wrong with her grandchildren, reported experiencing considerable levels of anxiety and worry following diagnosis:

"...It changed a lot of things. Because we were, I couldnae go to sleep at night because I was up checking her to see, because the Dr explained that they can go in their sleep- most of them go in their sleep when that, it does them over, and I'd just be up checking her to see they're, you know up, [] and if you felt they weren't breathing you were giving them a wee shake there." (Terri)

Terri tragically lost her daughter-in-law (the children's mother) to a cardiac arrest caused by undiagnosed LQTS, and this might have heightened the level of fear she experiences. Her sleep difficulties and worries have reportedly decreased since the diagnosis but are still present. Constant worry and anxiety were emotions mentioned by all of the caregivers as being part of their daily lives. Most of the caregivers regarded this constant worry as a burden; however Jennifer appeared to have accepted her worry as normal:

"Well, he was only 5 weeks old, so because of that it's been, our lives have always been just living with [son]'s cardiac arrhythmia you know. And, em he's been on medication his whole life- since he was 5 weeks old- so, it's totally normal for us (laughs)[] Everything we do and all the worries we have about him every day are just quite normal, as part of our lives. I think it might be more difficult if a child's older you know. " (Jennifer)

Jennifer reflects that they have found it easier to adapt to living with their son's condition because it has always been that way for them. Her son's condition has also been well managed by medication since diagnosis, which is not the case for all of the families interviewed. Jennifer's suggestion that it might be more difficult to adjust if your child is diagnosed when older seems to be true in the case of Mary, who stated the following:

"...When [son] got diagnosed with *that*, I thought my world was ending. Because everything, everything cha, *everything* changed. The way we did things as a family changed. So now we're doing things, well, not so much now, but then we were doing things differently." (Mary)

Mary's statement suggests that having to completely adjust family life post diagnosis made it difficult to adjust to the diagnosis itself. She implies that they have now adjusted as a family because their adapted activities are now the norm for them, indicating that the process of adjustment is dynamic and takes time.

Describing their process of adjustment from a more holistic perspective, Jane and Jack reported that after the initial shock of receiving their son's diagnosis, they realised that learning of his diagnosis was a gift:

"It's like what Jack's just said, is it's a *gift* to know your child's got a heart condition. And it took us maybe about a year to really truly accept that, didn't we? That Dr X and Dr Y had *gifted* us with knowledge. Had we had we not been in the UK, had the *privilege* of Yorkhill Hospital, had we not known or been reactive to [son]'s situation, [son] wouldn't be here, that's just a given. That that's just a *fact*." (Jane)

The experience of almost losing their child seemingly helped them realise how lucky they were that he was still alive. This was very similar to Jennifer's perception of her situation. These caregivers also reported that living with their child's condition had strengthened their family as a whole. Their terrifying early experiences appeared to have positively influenced their ability to cope with the diagnosis and see it in a positive way.

Another significant way in which the diagnosis affected the families whose children were now in their late teens was in terms of loss of identity and future career. Jane and Jack's son was a promising goalkeeper, and Jack described a considerable loss in relation to this:

"He'd gone to [anon football club] , was going to their training and performed *incredibly* in their training nights, a couple of training nights they had, and more or less they'd invited him up to [anon football club] to *play*, and were gonna sign him, more or less. And at that point, I said yip, I'm gonna be upfront here and declare he's got this condition, this eh heart condition. At which point, you know, they froze, 'eh, sorry, that wasn't quite what we mentioned, that wasn't quite what we meant', eh and they *didn't* take up the option for him you know, with that, because they said well, look with that heart condition, you're never gonna make it as a, as a, you know, as a professional footballer." (Jack)

This loss of opportunity for their son appeared to devastate Jack and Jane. Interestingly, there were differences in their emotional reaction to the situation: Jack felt strong empathy for his son, however was also able to understand it from the football club's position. Jane on the other hand openly recognised that she took the decision more personally. Jane portrayed her reaction powerfully in the following quotation:

"...it was almost like they had just *abandoned* [son]. You know, 'you've got the plague, we're not touching you'. And yet, and I found that just so *hurtful* for [son], because he was probably at the top of his game at that point, and you know, he's he's a robust wee boy- big boy- and that to me was just like, oh my *goodness*, you know, [son] is, I got all sort of, quite, em, I'm going to try and say this being as politically correct as I can, but, I thought...yeah they they done a wrong thing. They really done a *wrong* thing." (Jane)

Jane's perception that the club's view of her son's cardiac arrhythmia is akin to him having "the plague" suggests her belief that he is being unjustly stigmatised and discarded.

Mary reported that her son had always been physically active growing up, and described his emotional response to the diagnosis:

"[Son] at 11 was basically told he couldn't play sport. That was it. There's nothing, and that he could die. There was nothing else for [son] at that point. [Son] was *very* resentful. *Very* ang- *very* very angry." (Mary)

Additionally, Mary reported that her daughter was not a physically active person and therefore had not been immediately affected by the diagnosis in the same way as her son. It did affect her later on, however, when she realised that she would not be able to become a policewoman as she had always hoped:

"And to find out she couldn't be a policewoman, my daughter's devastated. That has really, and my daughter still talks about it." (Mary)

As can be seen, loss of identity and potential career was a significant factor for two of the families in the study. Given that the children in two of the other families were still very young, this may potentially become a factor for them in the future.

Living with Uncertainty

All of the caregivers described that one of the hardest parts of living with their child's condition was living with the uncertainty of what might happen on a daily basis and in the future. Grant introduces this well:

"The thing that's been hardest, I suppose for any parents to get their heads round is that there's a huge area with [son] that is *unknown*. There's a whole unkno- they don't *know*. They don't *know* quite why this is happening. They don't actually quite *know* why these drugs are working." (Grant)

Similarly, Terri provided a good description of the ways in which unexpected events can occur at any time and how this feels for her:

Terri: "Although they're getting the medication, like something could just go wrong. Like what happened with [granddaughter 1], she was on the medication and she was taking wee turns. And the Dr had to give her another pill on top of that. So you know these things come up.

Interviewer: So almost like trial and error in some ways?

Terri: Yeah, and if they get a fright or fall into cold water the shock could kill them, you know what I mean?

Interviewer: OK, yeah.

Terri: It's quite frightening actually."

Finally, Mary provided a perceptive metaphor to conceptualise how it feels for her and her family:

"I feel we don't get answers- but that's not [cardiologist]'s fault. That's because the condition can't give you answers. Cause it's a very, the way I look at it, sometimes it's like an unexploded bomb, it's like a ticking time bomb. It could be in the ground for years and nothing happens. [Son] could live till he's a hundred and nothing'll ever happen to him! But there's always that chance. He's got more of a risk than somebody else. And that's, as a family that was the hardest thing to deal with." (Mary)

Mary's description provides a valuable insight into what it must feel like every day for these families. Her use of the phrase "a ticking time bomb" emphasises just how serious the outcome could be for her children if something was to go wrong and how little control she has over the likelihood of this occurring.

Balancing Risk with their Child's Wishes

Another way in which the cardiac arrhythmia affected the caregivers' daily lives was in forcing them to constantly balance the risk of a situation with their child's desires to engage in that situation. This had been a clear part of the narrative for Jack and Jane throughout their son's life, and it was apparent that it was an on-going challenge for them even with their son now aged 17 years old. They reported that he was currently considering taking up rugby, something they had encouraged him to refrain from in the past when they still had legal parental responsibility for him:

Jack: "Do you know and if he decides that he wants to go and take up rugby, and he signs the papers that say, look I've got the heart condition, then that's up to him, d'you know, we'll go along and

support him, that's his *choice*, but em I'm hoping he doesn't! But that, but that's that's that'll be his, and again it's just, our attitude was, it's not going to hold him back, we want him to go out, *live* his life to the full, what's the point in living a life-

Jane: What's the point in having a life if you don't live it?

Jack: Yeah. And so he's got that sort of mentality thing."

It appears that Jack and Jane's philosophy on life is part of what helps them manage their worries regarding their son engaging in high-risk activities. Terri described a similar philosophy and reflected on how she dealt with her anxieties about what might happen to her granddaughters:

"You've just, you've got to let them live their life, you cannae just keep them wrapped up in cotton wool all the time. You know what I mean? Don't let, especially when they're teenagers, you're frightened if anything happens but you've got to, you just cannae keep them tied up. You know what I mean. It's their life." (Terri)

Mary described that they, as a family, had developed their own method of risk assessment for her children, in which they had agreed on certain rules for what they could and could not do:

"As a family we, we've sort of risk element-wise, he doesn't jump in a swimming pool, never has. He walks into a swimming pool. Physical exercise: he doesn't play competitive football anymore. Rugby: didn't play competitive rugby. " (Mary)

The challenge of balancing risk with their child's wishes appeared to be especially difficult for caregivers with children in their teenage years, due to teenagers' increasing desire for independence, autonomy and to engage in new and exciting activities with peers.

Experiences of Living with the AED

All families had been given an AED in order to maximise the chance of their child surviving in the event of him/her experiencing a sudden cardiac arrest. Three main

themes were revealed across the caregivers' reports regarding this: the impact of being given the AED; using the AED; and the AED and school.

The Impact of being given the AED

Four of the families discussed how it felt to be given the AED initially. Caregivers' feelings in response to being asked to take the AED home differed within the sample. Lisa described her and Grant's initial response:

Lisa: "It was a scary thing for us at first, to be told, you know, we suggest you take this home. We were like 'oh, my god!' you know?"

Interviewer: What do you think, what was it about it that was a scary thing?

Lisa: Well, the reason you have to have the defibrillator is because if their heart stops...you can get it going again. And that's, that's quite hard to hear. Especially when you're living with him day to day and he's- see, as far as we've always been concerned, when he's on his medication and it *works*, then we've always felt there's no reason for him to have a collapse."

Lisa's reflection shows that being given the AED served as a reminder of the probable fragility of their son's life and contradicts their perception that their son's health is under control. This is similar to Jennifer's response:

Interviewer: "I mean how did it feel for you to be given a defibrillator to take away with you?

Jennifer: Well it felt really really strange. That that *still* feels odd that we have that in the boot of our car all the time. So we've never had to use it, fortunately. You know, em, it's just, yeah it's a bit of a scary reminder every time I see it in fact. Whereas everything else is quite normal, you know giving him his medication and all that stuff is totally normal and it doesn't phase us at all, but I suppose the thought of having to use that, yeah that's the scariest thing of all probably."

Jennifer also perceives that her son's health is under control and sees being given the AED as a contradiction of that belief, which she finds very unsettling.

Despite communicating anxiety regarding the thought of having to use the AED, both Terri and Mary expressed a different reaction to being given the AED, namely one of relief. This is portrayed well by Mary:

Interviewer: “And how, how did you feel at the time when they offered you the defibrillator? What was your reaction?”

Mary: Oh, relief. Relieved.

Interviewer: Relief.

Mary: Relieved. Because I knew if anything happened to him, that I was quicker than going to hospital. It was a, it was there...”

Mary felt relief and comfort at having the AED. This echoes Terri’s feelings about it, who reported that the AED had become “part of daily life” for her and her family.

Using the AED

None of the caregivers had needed to use the AED on their child at the time of being interviewed. Nonetheless, the previous episodes suffered by some of the children had given some of the caregivers a good insight into what it might feel like for them if they were required to use the AED in future, and this affected their perception of their ability to use the AED. Jane compellingly described her reflections regarding this:

“But *actually* when the episodes occurred, even as parents, even though your *complete* instinct is to keep your child alive no matter what the scenario, when adrenaline kicks into your own body it is hard to remember... cause all you want to do is kinda scream and hold your baby, and, bring that baby back to life just by sheer *will power* rather than actually any actions that you might *have* to do. So it does take an amount of *discipline* back into your own self to then administer the structured path that, you know, that brings your child back. So that that was a kind of a big step to get over, eh and just to understand your own emotions that are part of that... ” (Jane)

Here, Jane describes the impact of anxiety on her ability to function in past times of crisis, and the way this became a barrier for her to overcome. Lisa also voiced concerns about the effect that anxiety might have on her ability to use the AED:

“It’s obviously different to go through all that scenario with somebody that’s lying on the street rather than your own child. Do you know what I mean, the panic that comes into you when it’s your child rather than, you know, even another family member or whatever, it is, it’s totally, totally different.” (Lisa)

There appears to be an important distinction for Lisa and Jane between administering the AED on their own child and administering it on someone else. This emphasises how difficult it was for them to manage their distress in the past when their child was experiencing a seizure. Their past experiences intensify their worries regarding administering the AED on their child should the need arise. Terri did not appear as concerned about her ability to use the AED despite voicing initial anxiety about the thought of having to use it:

“I think I would...I think my mind would make me do it, you know, I’d have to do it for the kids, you know. Hopefully I could do it then, but I think I would be able to do it no bother.” (Terri)

Only one of the caregivers (Jennifer) reported having received refresher training in using the AED since being given it, and she reported this to have been very helpful. The four other families reported that they would like refresher training and felt this would increase their confidence in their abilities to use it.

The AED and School

Three out of the five families reported experiencing difficulties with obtaining an AED for their child in school despite the hospital recommending this. Jennifer was the only caregiver who reported no difficulties with school, and that was possibly because the school already had an AED in place. There appeared to be wide variation in the support offered between the primary and secondary schools, with primary school seemingly being more supportive than secondary school in these three cases. Jane described that the transition from primary to secondary school had been “quite traumatic” for her and her son despite attending numerous meetings to put the

necessary measures in place prior to his transition. She described an on-going argument between the school and the council over who should fund the purchase of the AED, leading eventually to her and Jack offering to purchase the AED themselves. Jane described what it was like when her son started secondary school and there was no AED in place:

“I was asked to come to lots of meetings and then at one point it got to the stage where they were like, well [son] can’t do PE, [son] can’t do home economics, and there was something else- woodwork, or anything with a machine, in case he had a seizure when he was using it... And I kinda sat there thinking this is my son you’re talking about and yet it’s a bit of a rammy. So that *wasn’t* a pleasant experience, although in the end they were fantastic and they *have been* fantastic.” (Jane)

Fortunately the school eventually purchased an AED and, as expressed by Jane, have been supportive since her son’s difficult transition. A similar experience was reported by Mary:

“Regarding school, I had asked about a defibrillator: they weren’t going to do it. They weren’t going to give us.... [son]’s school weren’t going to give us it. And they’re no, we weren’t getting it. So I said, no, [son]’s getting it, I said I don’t care how you get it, just don’t buy a chair. You get it. I said, but, and the way I looked at it is, I had said if anything happens to [son], I said, and you don’t have one, I said I’ll be asking questions. [Son]’s defibrillator was bought- it was actually bought through a charity organisation...” (Mary)

It is apparent that the schools only fulfilled their obligations to Jane and Mary after the mothers engaged in several meetings and active assertive dialogue regarding the importance of the school having an AED. Unfortunately Terri struggled to adopt this assertive role with staff at her granddaughters’ school, and they have still not purchased an AED despite promising to do so six years ago when the eldest started attending. Terri reflected that she had not followed this issue up with school staff and had not mentioned it to the hospital team, due to being of the understanding that it was the school’s responsibility to follow it up. It appears that Terri found it difficult to adopt the proactive role assumed by Jane and Mary, and that the school subsequently did not fulfil their promise to her family. This emphasises the

vulnerability of some families who might lack the personal resources to pursue such important issues with external agencies.

Although Lisa and Grant had been advised by their Consultant Cardiologist to ensure their son's primary school obtained an AED, and they reported that the school had been supportive of their son's needs, they had not yet informed the school:

"...they (the cardiologist team) wanted the school to have a defibrillator as well, and we, we kind of, we weren't we're not against that, but we said well, let's just see how we get on with it first. And, because we didn't want to put the fear into the school because, em, I was glad when they gave it to us, and I was, you know I was happy to take it. But, sort of in the back of my mind I was kind of like 'I'm not sure if this is necessary for him'." (Lisa)

It appears that Lisa and Grant might not be fully aware of the role and function of the AED and might not understand the importance of it in the event of their son experiencing a cardiac arrest. Lisa also described at another point in the interview that their AED had been kept in a cupboard since the day they brought it home and "hasn't been taken out since". This indicates a clear need for health professionals to ensure caregivers fully understand the role and function of the AED in relation to their child's condition, and clearly communicate the reasons for advising that school obtain an AED. It appears that facilitating communication with school regarding the importance of the AED would also be helpful for many families.

Experience of Hospital Support

"All the things that the medical team put in place here, the consultations, the regular check-ups, the information, the em the genetic stuff of going and searching for that, all the things that they did, em put us in a better place to deal with it and manage it as a family. And so you know, looking back on the whole experience, would we be where we are today without everything that had been done? Maybe not. You know, so it was a whole raft of things. [] At each stage, there was help provided and information came there that allowed us to manage it." (Jack)

This quotation by Jack is representative of the expressed opinion of all of the caregivers; they all reflected on the support they had received from the hospital since diagnosis, and praised the hospital staff for their dedication and commitment in

treating their child's condition. Two main themes emerged in relation to the caregivers' experience with hospital support: the small things make a big difference; and communication. These will each be discussed.

The Small Things Make a Big Difference

All of the families discussed various ways in which their experience of hospital support had been valuable in helping them manage their situation. It became clear through the interviews that the small things were very important in giving the families a positive experience. One example of this was given by Terri, who stated that her three granddaughters were always given cardiac appointments for the same date:

“ ‘Cause it was good when we, when I came up the first times, the doctor made sure the three of them were together, so just coming up once. So the three have always come up together which was, which was an awfa difference to us you know what I mean.” (Terri)

Similarly, Mary reflected on how important the small things were, and this had become more apparent to her since her son had transitioned to adult services:

Mary: “I always had a contact. The [] cardiologist clerk or whatever- amazing. Cause sometimes I have to phone up and say can you get me a letter for, for example I'm going on holiday, I'm going on the plane, we need a letter for the defib. Might not ask for it, but you need a letter. Do that no problem. So I've got a contact. I always felt if I ever needed Yorkhill- that's one thing I will say- I ever had a sort of issue or whatever, cardiac liaison nurse was brilliant, she would always get back to you. And the clerical. Whereas I don't feel like I've got anyone...

Interviewer: So it's not the same type of support network [in the adult hospital]?

Mary: Not at all. Just for stupid things. Do you know what I mean, just for stupid things.”

On a less positive note, however, Mary reported how frightening it had been for her son to transition to adult services, and she felt strongly that the hospital should have provided some support and preparation with his transition:

"I do think they should have a transition. It's not good enough they don't. Cause it's scary. It's scary..." (Mary)

Jennifer stated that she always felt "safe" in Yorkhill Hospital and continually praised the staff team:

"I've just always found them amazing in the ward. All the staff, just the way, they're just great, and it's really good that it's the same people. And we've had the same consultant the whole time which helps as well. So, and and it never, em ceases to amaze me when we go down and they remember, they remember us and they remember him and they must have seen hundreds of people since they've seen us last!" (Jennifer)

Finally, Lisa and Grant discussed that being given a stethoscope to monitor their son's heart rate at home had been incredibly empowering for them, and they felt that this was something all parents of children with a heart condition should be given:

Lisa: "It's so that parents feel they can do something themselves- I think that's the thing, I think that parents have to feel that they can *do* something themselves.

Grant: Yeah.

Lisa: Because when you go home, you are, you're pretty helpless. You're relying on the medication to work, em, you know. It's nice to be able to hear, or to be able to kinda go well 'do this and if it sounds fine, he's fine' and then that's you. Great!

Grant: You have no idea how wonderful it was to get a stethoscope. It was! It felt great."

Being given a stethoscope gave Lisa and Grant the sense that they could actually do something in a situation where they had little perceived control otherwise.

Communication

Despite all of the positive things said about their experience of hospital support, there were also some issues raised by two families regarding incidents they had found difficult to manage as a result of communication from hospital staff. These

difficulties were mainly expressed by Mary, and Lisa and Grant, however given the apparent significance of these communications for their experience of hospital support it was felt that it was important to include them. This is in the hope that it may serve as helpful information for hospital staff working with this population in the future.

First of all, Lisa and Grant expressed that they often found communication from hospital staff challenging, as they felt that sometimes staff would make offhand comments that would be deeply hurtful or distressing for them to hear. Lisa described this well:

“...You hang on to *every* word, good or bad. I think you hang on to more bad stuff than good stuff. ‘Cause if they say, he’s doing great, you kind of go, ‘pew’, fine! But see if they say ‘oh we don’t know, it’s not what we want’, you’re suddenly: ‘what do you mean it’s not what you want? Is it really bad?’ You know, you do, you get like you’re almost grabbing them and saying, and sometimes I think they maybe forget that, so they just rhyme off well ‘d-d-d-d’, you know.” (Lisa)

Lisa and Grant additionally reported feeling it was important to be treated as equal partners by the professionals, and stated that sometimes they didn’t feel as if they were treated in that way:

“There kind of has to be respect from both sides, and sometimes sometimes you feel as if, as a parent, you don’t get it.” (Lisa)

There was a clear sense that Lisa and Grant often did not feel heard by professionals within the hospital, and felt that their opinion should be taken into consideration more readily than it was. This may be linked to their belief that they are the experts on their child’s condition:

“I think one of the things maybe the the doctors are maybe a wee bit blasé about is, we’ve got to live with him every day. We know him better than anyone else. And I totally get that they are the experts, they know the medications, what they do, d-d-d-d-da, but on a daily basis, no one knows him better than us.” (Lisa)

Mary also had similar difficult experiences with communication from the medical professionals. The first difficult experience related to the way in which her son was informed of his diagnosis. She reported that her son at 11 years old was told that “he could die”, and strongly felt that this was not appropriate and may have negatively affected his adjustment to the condition as a result:

“I’d have preferred for them to have not mentioned that in front of [son]. I think sometimes there’s a need to know. And I don’t think at 11 years old when you’ve been running about doing things you need to be told that. Yes, you need to be told that there is certain things that if, you shouldn’t do to like exacerbate it like don’t do things like that. [] But I’m just saying from a point of view of telling an 11 year old they could die- no I didn’t think they did it the right way and I don’t think we got any support whatsoever after it.” (Mary)

As can be seen here, Mary also reported that they were not offered any follow-up support and stated several times throughout the interview that she felt being offered some sort of mental health support directly following diagnosis would have made a big difference to her son’s adjustment. Another challenge Mary had encountered more recently involved communication regarding the AED at the time of transition:

Mary: “At 18, I thought it was a bit heartless, first thing they- I hadn’t even actually had an appointment for the Royal, and I was asked for the defibrillator back...I was *asked* for the defibrillator *back*.”

Interviewer: From Yorkhill?

Mary: from Yorkhill. Hadn’t even hut- you’re literally talking weeks he’d left, and it was like right, give me the defibrillator back. And I said no, because I still had [daughter]. And I said n-nooooo, I says I’m keeping it until [daughter]’s 18. [] It’s as if, as soon as you hit that age, you’re gone. And anything you’ve got belonging to us we want back. I mean to ask for it that quick I thought was quite heartless, it really was.”

When considering the significant role and function the AED has served in her son’s life since his diagnosis age 11, Mary’s shock at this unexpected recall is understandable.

DISCUSSION

This study explored the lived experience of caregivers caring for children with cardiac arrhythmia who have an AED. Three super-ordinate themes have been reported on here: impact of cardiac arrhythmia on daily life, experiences of living with the AED, and experience of hospital support.

Impact of cardiac arrhythmia on daily life

The child's early seizures seemingly had a substantial impact on caregivers' emotional responses and adjustment. Those who had witnessed their child suffer an undiagnosed seizure or cardiac arrest remembered this vividly and subsequently frequently reminded themselves of how lucky they were that their child was still alive. These caregivers also felt that living with their child's condition had strengthened their family as a whole. It appears that experiencing a traumatic episode may have served as a catalyst for these caregivers to positively reframe their experience. This positive reframing functioned as a coping strategy, helping them to manage the uncertainty of their child's condition by looking for the positives in their situation, re-evaluating their priorities and values in life, and reminding themselves of how much worse their situation could have been. This is similar to the coping strategies reported by some parents of children with other chronic illnesses (*cf.* McNeill, 2004; O'Brien, 2001; Sullivan-Bolyai et al., 2006). Folkman and Moskowitz (2000) propose that positive reappraisal is particularly important in helping people to sustain their efforts in activities such as caregiving over long periods of time, and this provides support to the researcher's clinical observation that these specific caregivers appeared to be coping remarkably well with sustaining their caregiving role.

Interestingly, caregivers in this study who had not witnessed such a traumatic episode did not appear to use positive reappraisal to the same degree, and appeared to focus mainly on the negative impact of the condition. These latter mentioned caregivers tended to employ problem-focused coping strategies such as seeking information and acquiring helpful resources. This was a common coping strategy used by caregivers of people with AIDS in a longitudinal study by Moskowitz et al (1996) and has been identified as a helpful coping strategy in previous literature (e.g. Lazarus & Folkman, 1984). At the time of the research by

Moskowitz et al., little could be done to predict or control the course of AIDS, similar to the situation of the current caregivers. They found that caregivers pursued realistic goals by focusing on specific, small tasks, in order to retain some control in an uncontrollable situation. The current study revealed similar findings.

The age of the child at diagnosis appeared influential in how it affected the caregivers and the family. For example, Jennifer stated that because they received the diagnosis when her son was a baby, they adjusted easily to it. Mary on the other hand, whose son was diagnosed age 11, reported that the whole family struggled to adjust following diagnosis, especially her son. Additionally, there was a sense of loss and injustice for some of the older children when the diagnosis prevented them from pursuing a goal or career choice. The idea that diagnosis may be most disruptive to an individual's development during their adolescence and teenage years can be understood in terms of Erikson's (1963, 1968) psychosocial theory of development, which proposes that children must develop a sense of industry and competence in the years leading up to adolescence, and that during adolescence their task is to find their identity and role in the social world. Failure to achieve these tasks successfully, for example due to chronic illness, can lead to feelings of inferiority and role confusion for the adolescent (Erikson, 1963, 1968). Caregivers' reports that the diagnosis and seizures caused disruption to the family as a whole can be contextualised in terms of family life cycle theory (Hoffman, 1980), and the fact that the cardiac arrhythmia may have caused complications and disruptions to the traditional stages of the family life cycle (Rolland, 2010). According to family life cycle theory, periods of transition are potentially the most vulnerable (Hoffman, 1980). If chronic illness coincides with a period of transition such as adolescence, issues related to previous, on-going and anticipated loss will be amplified (Rolland, 2010). This provides a framework to understand why diagnosis was particularly difficult for certain families in the current study.

Importantly, Mary stated that she felt it should be mandatory that families are offered follow-up mental health support after diagnosis. She believed the lack of support her family received post-diagnosis compounded the negative impact of the diagnosis on her son's wellbeing and adjustment. The finding that receiving a diagnosis of cardiac arrhythmia in adolescence can have a particularly negative impact on the child and

family is supported by the study by Farnsworth et al. (2006). They recommended that close follow-up of adolescent patients following diagnosis is crucial to ensure positive adjustment to the condition. Given the current findings, this recommendation would seem very appropriate. Additionally, providing psycho-education to caregivers regarding the impact of chronic illness on the child and family's development in the context of Erikson's (1963, 1968) theory and the family life cycle theory would help them prepare for and manage any difficulties encountered.

All of the families reported that one of the hardest parts of living with their child's condition was living with uncertainty on a daily basis. This seemed to be an on-going challenge. Living with uncertainty is a familiar feature of families living with paediatric chronic illness (*cf.* Anker-Petersen, 2014 and Farnsworth et al., 2006) and has been consistently associated with negative psychosocial adjustment in parents and children (Fedele et al., 2011). It appeared that caring for children in their teens increased uncertainty for caregivers in this study, due to the child's increasing desire for independence and to engage in "high-risk" activities with peers. This supports research by Fedele et al. (2011) who examined the association between illness uncertainty and parent and youth adjustment in Juvenile Rheumatic Diseases. Interestingly, Farnsworth et al. (2006) conclude that uncertainty does not seem to be an on-going or pervasive emotion in the experience of parents of children with LQTS. This is at odds with the current findings. A significant limitation to the study by Farnsworth et al. is that it was based on short written survey responses as opposed to in-depth semi-structured interviews, and this may play a role in the differing conclusions reached.

Given the negative impact of uncertainty on psychosocial adjustment, it is important that medical staff take measures to reduce parental uncertainty. This can be done through providing clear, basic information to caregivers when possible, discussing and encouraging problem-solving skills with caregivers, and increasing open communication with medical staff (Fedele et al., 2011). Supporting caregivers to balance their child's increasing desire for autonomy with the management of their condition would be important. Additionally, one family emphasised how empowering being given a stethoscope had been. If resources allow, this might be a helpful and

relatively simple way of empowering all caregivers of children with this high-risk category of cardiac arrhythmia.

Experiences of living with the AED

No prior qualitative research has been undertaken, to the author's knowledge, on the experience of living with an AED when caring for a child with cardiac arrhythmia. This study found that some caregivers experienced high anxiety levels and fear in relation to the prospect of having to use the AED on their child, and some expressed concern regarding the impact of anxiety on their ability to use it. Other caregivers reported feeling relief at having the AED. Interestingly, Rahman et al. (2011) found that parents of adolescents with an ICD consistently reported feeling reassured by the ICD, and some reported giving their child greater independence and freedom as a result of the ICD. One adolescent in the study specifically reported experiencing a greater sense of freedom as a result of the ICD in comparison to the AED she previously carried. This indicates that the high level of caregiver responsibility associated with having an AED (as opposed to having an ICD where caregiver responsibility is essentially removed) may considerably increase caregiver anxiety levels and fears in relation to their child's condition, and may impact on the amount of independence and freedom they feel able to allow their child. Additionally, two caregivers in this study reflected that the experience of being given the AED was considerably more anxiety provoking due to it being in relation to their child as opposed to someone else. The nature of relationship between AED operator and patient has been noted as a concern in the adult literature when considering the potential psychological impact of placing AEDs in the homes of at-risk patients (Cagle et al., 2007). Findings have been mixed regarding the psychological impact on spouses of having an AED in the home, with some studies finding higher anxiety levels in spouses and others finding feelings of increased security and control for spouses (Cagle et al., 2007; Chen et al., 2002; Thomas et al., 2010). This is similar to the mixed picture gained in the current study and indicates that individual personality variables may play a role.

Thomas et al. (2011) found higher long-term anxiety levels in spouses who had been trained to use an AED in addition to CPR as opposed to spouses who had only

received CPR training. This led them to recommend an assessment of spousal anxiety levels in any family who had been given an AED, and appropriate follow-up support with anxiety management strategies if required. The current study would support the application of this recommendation in relation to caregivers of children with cardiac arrhythmia who have an AED.

Most of the caregivers reported low confidence levels in their ability to use the AED if necessary. Four out of the five families reported that they had not received refresher training since they had initially been given the AED. They all felt refresher training would be helpful in terms of improving their confidence levels and ability to carry out the steps correctly. Interestingly, the caregiver who had received frequent refresher training did not report low confidence levels in the same way as the others. This supports the suggestion that refresher training would be helpful for these families and is consonant with the proposal that problem-focused coping strategies increase feelings of efficacy and mastery in situations where people have little control (Folkman and Moskowitz, 2000). Cagle and colleagues (2007) recommended that AED training for spouses should include supporting the family to incorporate the machine into daily life. This would also be helpful for the current population. This could be done via a home visit to the family after initial training, to assess their adaptation to the device and provide appropriate follow-up support as required.

This study found that several parents had experienced significant challenges with ensuring their child's school obtained an AED, despite medical recommendations regarding the importance of this. Additionally, one couple did not seem to understand the need for the school to have an AED despite receiving explicit medical instruction. This is concerning considering the function of the AED and the associated increased risk of death for the child if they were to experience a cardiac arrest in school with no AED available. This indicates the need for better education and training of school staff regarding the risks of cardiac arrhythmia and the function of the AED. Additionally, on-going monitoring of caregivers' perceptions of their child's condition and their understanding of the requirement of the AED would be helpful. It is recommended that this issue is seriously considered when planning future service development for supporting this population.

Experience of Hospital Support

All of the caregivers in this study were, overall, very positive regarding their experience of hospital support and were full of praise for the staff team at Yorkhill Hospital. Two families had unfortunately experienced some difficult interactions with hospital staff, and this was seemingly due to difficulties in communication between staff and caregivers. A large body of research has explored the area of communication between medical professionals and patients, especially in relation to communicating difficult news. It is clear that an insensitive approach increases distress in patients and may exert a long-term negative impact on their ability to adjust (See Fallowfield & Jenkins, 2004, for a review).

The difficulties reported in the current study mainly related to the wording sometimes used by medical staff when providing feedback about the child's condition and treatment (similar difficulties were reported by parents in the study by Rahman et al., 2011); the way in which the diagnosis was given; and lack of preparation and support with the transition from child to adult services, also in relation to the AED. Additionally, one family reported that they felt it was important to be regarded as equal partners in their child's medical care and felt they were not treated in this way at the moment. Considering these difficulties, recommendations for improvement in the hospital support for this population include: ensuring communication is sensitive and considerate of the child and family's needs at all times (See Fallowfield and Jenkins, 2004, for a review of treatment guidelines and training available regarding this); ensuring the child and caregivers are supported in their transition from child to adult services; and ensuring appropriate information and preparation is given regarding the recall of the AED at the point of transition.

Strengths and Limitations

This research provided an insight into caregivers' experiences of caring for a child with cardiac arrhythmia who has an AED, which will be disseminated to practitioners working with this population. A greater understanding of caregivers' experiences can be used to inform services about how they can best support and meet the needs of children with cardiac arrhythmia and their families, and several recommendations for future practice and service development have been proposed here. A Paediatric Clinical Psychology service has recently been established within the RHSC

cardiology department, and this research will help directly inform the new service development.

There are only 16 families in Scotland who currently have a child with cardiac arrhythmia and who have an AED. Therefore this research has explored the experiences of almost a third of the targeted population through the use of in-depth, semi-structured interviews. Everyone in this population in Scotland was invited to take part in the research, and therefore the researcher could not have obtained a larger sample size unless she had recruited in other countries too, which was out with the scope of the current study. A small sample size is well-suited to IPA, which maintains an idiographic focus throughout all stages of analysis. The researcher tried to stay true to the philosophy of IPA throughout all stages of the research process, and this can be shown via the clear audit trail that exists from all stages of her research. Please contact the researcher for further evidence relating to the audit trail if this is required. Additionally, the researcher used the IPA guidelines developed by Smith (2011) when writing up this paper, in order to hopefully achieve what would be considered a “good” IPA paper.

One of the interviews carried out in this study was a telephone interview. It is recognised that a telephone interview may make it more difficult for the interviewer to establish rapport with the participant, and will prevent the interviewer from observing the participant’s non-verbal communication which can provide a context for subsequent analysis (Turner, Barlow & Ilbery, 2002). It was felt, however, that conducting a telephone interview was a superior option to denying Jennifer the chance to discuss her experiences. Reassuringly, the use of the telephone did not appear to disrupt rapport building in this interview, and Jennifer informed the researcher afterwards that she had found the interview therapeutic, evidence that a good relationship had been established between interviewer and participant.

A final noteworthy issue is that two of the interviews undertaken in this study were joint interviews. Both parents identified themselves as being a main caregiver, and asked to be interviewed together. Given that IPA focuses on understanding a person’s experience from their perspective, the researcher felt that it was important to respect the caregivers’ requests and allow them to discuss their experiences in a

joint interview. There are advantages and disadvantages to joint interviewing (see Arksey, 1996). Importantly, the researcher did not feel that the data was compromised due to having undertaking joint interviews. Both parents in both interviews contributed in almost equal amounts, and it allowed the researcher to observe the interactions between the parents which added context to the data. The main challenge in relation to this was transcribing the joint interviews. Overall, the researcher felt that the interviews generated rich and comprehensive data which contributed substantially to the findings of the overall study.

CONCLUSION

This study has provided a rich insight into the experiences of caregivers caring for a child with cardiac arrhythmia who has an AED. It has provided new data regarding caregivers' experiences with the AED, an area that has not been researched in the paediatric qualitative literature before. Several implications and recommendations for future practice and service development have been outlined (see appendix 15 for a summary of these), and this will be disseminated directly to practitioners working with this population.

REFERENCES

American Heart Association (2013). What is an Arrhythmia? [Online] Available at: <http://www.heart.org/HEARTORG/Conditions/Arrhythmia> [Accessed 21 January 2013].

Andersen, J., Oyen, N., Bjorvatn, C. & Gjengedal, E. (2008). Living with Long QT Syndrome: A Qualitative Study of Coping with Increased Risk of Sudden Cardiac Death. *Journal of Genetic Counselling*, 17, 489-498.

Anker-Petersen, S. (2014). Parental experiences of caring for a child with chronic illness: A meta-ethnography. *Submitted as part of the Clinical Research Portfolio Vol I*. Glasgow: University of Glasgow, 5-38.

Arksey, H. (1996). Collecting data through joint interviews. *Social Research Update, Issue 15*. Department of Sociology: University of Surrey.

Brown, R., Wiener, L. Kupst, M., Brennan, T., Behrman, R., Compas, B., Elkin, D., Fairclough, D., Friebert, S., Katz, E., Kazak, A., Madan-Swain, A., Mansfield, N., Mullins, L., Noll, R., Patenaude, F., Phipps, S., Sahler, O., Sourkes, B. & Zeltzer, L. (2008). Single Parenting and Children with Chronic Illness: An Understudied Phenomenon. *Journal of Pediatric Psychology*, 33(4), 408–421.

Cagle, A.J., Diehr, P., Meischke, H., Rea, T., Olsen, J., Rodrigues, D., Yakovlevitch, M., Amidon, T. & Eisenberg, M. (2007). Psychological and social impacts of automated external defibrillators (AEDs) in the home. *Resuscitation*, 74, 432-438.

Chen, M.A., Eisenberg, M.S. & Meischke, H. (2002). Impact of in-home defibrillators on postmyocardial infarction patients and their significant others; an interview study. *Heart and Lung: The Journal of Acute and Critical Care*, 31(3), 173-185

DeMaso, R., Lauretti, A., Spieth, L., Van Der Feen, J., Jay, K., Gauvreau, K., Walsh, E. & Berul, C. (2004). Psychosocial factors and quality of life in children and adolescents with implantable cardioverter-defibrillators. *The American Journal of Cardiology*, 93, 582- 587.

Department of Health (2005) National Services Framework for Coronary Heart Disease. Chapter Eight: Arrhythmias and Sudden Cardiac Death. [Online] Available at: <http://www.c-r-y.org.uk/NSF%20Chapter%208.pdf> [Accessed 1 December 2012].

Divekar, A. & Soni, R. (2006). Successful parental use of an automated external defibrillator for an infant with long-QT Syndrome. *Pediatrics*, 118, e526- e529.

Doniger, S.J. & Sharieff, G.Q. (2006). Pediatric dysarrhythmias. *Pediatric Clinics of North America*, 53, 85-105.

Dunbar, S., Dougherty, C., Sears, S., Carroll, D., Goldstein, N., Mark, D., McDaniel, G., Pressler, S., Schron, E., Wang, P. & Zeigler, V. (2012). Educational and

Psychological Interventions to improve outcomes for recipients of implantable cardioverter defibrillators and their families. A scientific statement from the American heart association. *Circulation*, 126, 2146-2172.

Erikson, E.H. (1963). *Childhood and Society*. New York: Norton

Erikson, E.H. (1968). *Identity: Youth and Crisis*. New York: Norton

Fallowfield, L & Jenkins, V (2004) Communicating sad, bad and difficult news in medicine. *The Lancet*, 363, 312-319.

Farnsworth, M., Fosyth, D., Haglund, C. & Ackerman, M.J. (2006). When I go in to wake them...I wonder: Parental perceptions about congenital long QT syndrome. *Journal of the American Academy of Nurse Practitioners*, 18, 284-290.

Fedele, D.A., Ramsey, R.R., Ryan, J.L., Bonner, M.S., Mullins, L.L., Jarvis, J.N. & Chaney, J.M. (2011). The association of illness uncertainty to parent and youth adjustment in juvenile rheumatic diseases: Effect of youth age. *Journal of Developmental and Behavioral Pediatrics*, 32, 361-367.

Folkman, S. & Moskowitz, T. (2000). Positive affect and the other side of coping. *American Psychologist*, 55 (6), 647-654.

Hanash, C.R. & Crosson, J.E. (2010). Emergency diagnosis and management of pediatric arrhythmias. *Journal of Emergency, Trauma and Shock*, 3 (3), 251-260.

Haskell, S. & Atkins, D. (2010). Defibrillation in children. *Journal of Emergency, Trauma and Shock*, 3(3), 261-266.

Hoffman, L. (1980). The life cycle and discontinuous change. In E.A. Carter & M. McGoldrick (eds.), *The family life cycle: A framework for family therapy*. New York: Gardner Press.

Kaiser, K. (2009). Protecting respondent confidentiality in qualitative research. *Qualitative Health Research*, 19 (11), 1632-1641.

Lazarus, R.S. & Folkman, S. (1984). *Stress, appraisal and coping*. New York: Springer.

McNeill, T. (2004). Fathers' experience of parenting a child with juvenile rheumatoid arthritis. *Qualitative Health Research*, 14 (4), 526-545.

Moskowitz, J.T., Folkman, S., Collette, I., & Vittinghoff, E. (1996). Coping and mood during AIDS-related caregiving and bereavement. *Annals of Behavioural Medicine*, 18, 49-57.

O'Brien, M. (2001) Living in a house of cards: family experiences with long-term childhood technology dependence. *Journal of Pediatric Nursing*, 16 (1), 13-22.

- Rahman, B., Macciocca, I., Sahhar, M., Kamberi, S., Connell, V. & Duncan, R. (2011). Adolescents with Implantable Cardioverter Defibrillators: A Patient and Parent Perspective. *Pacing and Clinical Electrophysiology*, 35, 62-72.
- Rolland, J.S. (2010). Chronic Illness and the Family Life Cycle. In M. McGoldrick, N. Garcia-Preto, & E. Carter (eds.), *The Expanded Family Life Cycle: Family, and Social Perspectives*. 4th edition. Boston: Allyn & Bacon
- Samson, R.A., Berg, R.A., Bingham, R., Biarent, D., Coovadia, A., Hazinski, M.F., Hickey, R.W., Nadkarni, V., Nichol, G., Tibballs, J., Reis, A.G., Tse, S., Zideman, D., Potts, J., Uzark, K., Atkins, D. (2003). Use of Automated External Defibrillators for Children: An Update. An Advisory Statement From the Pediatric Advanced Life Support Task Force, International Liaison Committee on Resuscitation. *Circulation*, 107, 3250-3255.
- Shinebourne, P. (2011). The theoretical underpinnings of interpretative phenomenological analysis (IPA). *Existential Analysis*, 22 (1), 16-31.
- Smith, J.A (2011). Evaluating the contribution of interpretative phenomenological analysis. *Health Psychology Review*, 5 (1), 9-27.
- Smith, J.A., & Eatough, V. (2007). Interpretative Phenomenological Analysis. In Lyons, E., & Coyle, A., (Eds.) *Analysing Qualitative Data in Psychology* (Chapter 3). London: Sage Publications.
- Smith, J.A., Flowers, P. & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, method and research*. London: Sage Publications.
- Sullivan-Bolyai, S., Rosenberg, R., & Bayard, M. (2006). Fathers' reflections on parenting young children with type 1 diabetes. *American Journal of Maternal Child Nursing*, 31(1), 24-31.
- Thomas, S., Friedmann, E., Lee, H-J., Son, H. & Morton, P. (2011). Changes in anxiety and depression over 2 years in medically stable patients after myocardial infarction and their spouses in the Home Automatic External Defibrillator Trial (HAT): a longitudinal observational study. *Heart*, 97, 371-381.
- Turner, A, Barlow, J. & Ilbery, B. (2002). Play hurt, live hurt: living with and managing osteoarthritis from the perspective of ex-professional footballers. *Journal of Health Psychology*, 7, 285-301.

Chapter 3: Advanced Clinical Practice I- Reflective Critical Account Abstract

A Reflection on the Development of my Communication Skills throughout Clinical Psychology Training

Abstract

According to the British Psychological Society (2010), communication skills are the primary building blocks of a Clinical Psychologist's role. This reflective account details my reflections of the development of my communication skills with patients, carers and colleagues throughout my Clinical Psychology training. The introduction discusses the reasons that I chose this aspect of my learning to reflect upon, and the context within which I have developed my skills in this area. The Integrated Development Model (IDM; Stoltenberg, McNeill, and Delworth, 1998) of Supervision and Gibbs' (1988) model of Reflection are described in the introduction and are drawn upon to structure and guide the reflection. The reflection itself takes the reader through various learning experiences that I have had in relation to communicating with patients, carers and colleagues within the multi-disciplinary team, and my reflections in relation to these. Following this, I reflect upon the process of writing my reflection, and what this has helped me realise in relation to my development over the three year training period, and what I may do to continue my development in the future. I also consider the relevance of my reflection in terms of the wider profession of Clinical Psychology and the evolving roles that Clinical Psychologists must fill in order to achieve the aspirations of policy and professional drivers for higher quality and efficiency in health care.

Chapter 4: Advanced Clinical Practice II- Reflective Critical Account Abstract

A Reflection on the Experience of Undertaking Research within the Context of Clinical Psychology Training

Abstract

Planning and conducting research is one of the core competencies required of a Clinical Psychologist, and substantially contributes towards the role of the Clinical Psychologist as a scientist practitioner. In this reflective account, I reflect on my experiences of carrying out my major research project within the context of continuous evaluation and assessment of my clinical skills and research skills as part of my Clinical Psychology training. I reflect on the experiences I have found both valuable and challenging within this context, using Gibbs' (1988) model to structure my reflection throughout. Following this I reflect on how the process of writing the reflective account has helped me identify how I have developed as a reflective scientist practitioner over time. This leads me to consider my continuing professional development needs, and what I can do to continue to develop as a reflective scientist practitioner throughout my career post-qualification.

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Appendix 1. Notes to Authors: Preparation for Submission to the Journal of Pediatric Nursing

Article structure

Subdivision - unnumbered sections

Divide your article into clearly defined sections. Each subsection is given a brief heading. Each heading should appear on its own separate line. Subsections should be used as much as possible when cross-referencing text: refer to the subsection by heading as opposed to simply 'the text'.

Appendices

If there is more than one appendix, they should be identified as A, B, etc. Formulae and equations in appendices should be given separate numbering: Eq. (A.1), Eq. (A.2), etc.; in a subsequent appendix, Eq. (B.1) and so on. Similarly for tables and figures: Table A.1; Fig. A.1, etc.

Essential title page information

- **Title.** Concise and informative. Titles are often used in information-retrieval systems. Avoid abbreviations and formulae where possible.
- **Author names and affiliations.** Where the family name may be ambiguous (e.g., a double name), please indicate this clearly. Present the authors' affiliation addresses (where the actual work was done) below the names. Indicate all affiliations with a lower-case superscript letter immediately after the author's name and in front of the appropriate address. Provide the full postal address of each affiliation, including the country name and, if available, the e-mail address of each author.
- **Corresponding author.** Clearly indicate who will handle correspondence at all stages of refereeing and publication, also post-publication. **Ensure that phone numbers (with country and area code) are provided in addition to the e-mail address and the complete postal address. Contact details must be kept up to date by the corresponding author.**
- **Present/permanent address.** If an author has moved since the work described in the article was done, or was visiting at the time, a 'Present address' (or 'Permanent address') may be indicated as a footnote to that author's name. The address at which the author actually did the work must be retained as the main, affiliation address. Superscript Arabic numerals are used for such footnotes.

Abstract

A concise and factual abstract with fewer than 100 words is required. The abstract

should state briefly the purpose of the research, the principal results and major conclusions. An abstract is often presented separately from the article, so it must be able to stand alone. For this reason, References should be avoided. Also, non-standard or uncommon abbreviations should be avoided, but if essential they must be defined at their first mention in the abstract itself.

Abstract is to conform to the APA 6th edition guidelines.

Keywords

Immediately after the abstract, provide a maximum of 6 keywords, using American spelling and avoiding general and plural terms and multiple concepts (avoid, for example, 'and', 'of'). Be sparing with abbreviations: only abbreviations firmly established in the field may be eligible. These keywords will be used for indexing purposes.

Acknowledgments

Collate acknowledgments in a separate section on the title page. List here those individuals who provided help during the research (e.g., providing language help, writing assistance or proof reading the article, etc.).

References

Please ensure that every reference cited in the text is also present in the reference list (and vice versa). Unpublished results and personal communications are not recommended in the reference list, but may be mentioned in the text. If these references are included in the reference list they should follow the standard reference style of the journal and should include a substitution of the publication date with either 'Unpublished results' or 'Personal communication'. Citation of a reference as 'in press' implies that the item has been accepted for publication.

Reference links

Increased discoverability of research and high quality peer review are ensured by online links to the sources cited. In order to allow us to create links to abstracting and indexing services, such as Scopus, CrossRef and PubMed, please ensure that data provided in the references are correct. Please note that incorrect surnames, journal/book titles, publication year and pagination may prevent link creation. When copying references, please be careful as they may already contain errors. Use of the DOI is encouraged.

Web references

As a minimum, the full URL should be given and the date when the reference was

last accessed. Any further information, if known (DOI, author names, dates, reference to a source publication, etc.), should also be given. Web references can be listed separately (e.g., after the reference list) under a different heading if desired, or can be included in the reference list.

Reference formatting

There are no strict requirements on reference formatting at submission. References can be in any style or format as long as the style is consistent. Where applicable, author(s) name(s), journal title/book title, chapter title/article title, year of publication, volume number/book chapter and the pagination must be present. Use of DOI is highly encouraged. The reference style used by the journal will be applied to the accepted article by Elsevier at the proof stage. Note that missing data will be highlighted at proof stage for the author to correct.

Appendix 2: Quality Appraisal Framework (Adapted from Walsh & Downe, 2006, page 114-115)

Stages	Essential Criteria	Specific Prompts
Scope & Purpose	<p>1. Clear Statement of and rationale for research question/aims/ purposes</p> <p>2. Study thoroughly contextualised by existing literature</p>	<ul style="list-style-type: none"> • <i>Clarity of focus demonstrated</i> • <i>Explicit purpose given such as descriptive/ explanatory, intent, theory building, hypothesis testing</i> • <i>Link between research and existing knowledge demonstrated</i> • <i>Evidence of systematic approach to literature review, location of literature to contextualise the findings, or both</i>
Design	3. Method/ design apparent and consistent with research intent	<ul style="list-style-type: none"> • <i>Rationale given for use of qualitative design</i> • <i>Discussion of epistemological/ ontological grounding</i> • <i>Rational explored for scientific qualitative method (e.g. ethnography, grounded theory, phenomenology)</i> • <i>Discussion of why particular method chosen is most appropriate/ sensitive/ relevant for research question/ aims</i> • <i>Setting appropriate</i>

	4. Data collection strategy apparent and appropriate	<ul style="list-style-type: none"> • <i>Were data collection methods appropriate for type of data required and for specific qualitative method?</i> • <i>Were they likely to capture the complexity/ diversity of experience and illuminate context in sufficient detail?</i> • <i>Was triangulation of data sources used if appropriate?</i>
Sampling strategy	5. Sample and sampling method appropriate	<ul style="list-style-type: none"> • <i>Selection criteria detailed, and description of how sampling was undertaken</i> • <i>Justification of sampling strategy given</i> • <i>Thickness of description likely to be achieved from sampling</i> • <i>Any disparity between planned and actual sample explained</i>
Analysis	6. Analytic approach appropriate	<ul style="list-style-type: none"> • <i>Approach made explicit (e.g. thematic distillation, constant comparative method, grounded theory)</i> • <i>Was it appropriate for the qualitative method chosen?</i> • <i>Was data managed by software package or by hand and why?</i> • <i>Discussion of how coding system/ conceptual</i>

		<p><i>frameworks evolved</i></p> <ul style="list-style-type: none"> • <i>How was context of data retained during analysis?</i> • <i>Evidence that the subjective meanings of participants were portrayed</i> • <i>Evidence of more than one researcher involved in stages if appropriate to epistemological/ theoretical stance</i> • <i>Did research participants have any involvement in analysis (e.g. member checking)</i> • <i>Evidence provided that data reached saturation or discussion/rationale if it did not</i> • <i>Evidence that deviant data was sought or discussion/ rationale if it was not</i>
Interpretation	<p>7. Context described and taken account of in interpretation</p> <p>8. Clear audit trail given</p> <p>9. Data used to support</p>	<ul style="list-style-type: none"> • <i>Description of social/ physical and interpersonal contexts of data collection</i> • <i>Evidence that researcher spent time 'dwelling with the data', interrogating it for competing/ alternative explanations of phenomena</i> • <i>Sufficient discussion of research process such that others can follow 'decision trail'</i> • <i>Extensive use of field notes entries/ verbatim</i>

	interpretation	<i>interview quotes in discussion of findings</i> <ul style="list-style-type: none"> • <i>Clear exposition of how interpretation led to conclusions</i>
Reflexivity	10. Researcher reflexivity demonstrated	<ul style="list-style-type: none"> • <i>Discussion of relationship between researcher and participants during fieldwork</i> • <i>Demonstration of researcher's influence on stages of research process</i> • <i>Evidence of self- awareness/ insight</i> • <i>Documentation of effects of the research on researcher</i> • <i>Evidence of how problems /complications met were dealt with</i>
Ethical dimensions	11. Demonstration of sensitivity to ethical concerns	<ul style="list-style-type: none"> • <i>Ethical committee approval granted</i> • <i>Clear commitment to integrity, honesty, transparency, equality and mutual respect in relationships with participants</i> • <i>Evidence of fair dealing with all research participants</i> • <i>Recording of dilemmas met and how resolved in relation to ethical issues</i> • <i>Documentation of how autonomy, consent, confidentiality and anonymity were managed</i>
Relevance & transferability	12. Relevance and transferability evident	<ul style="list-style-type: none"> • <i>Sufficient evidence for typicality specificity to be assessed</i>

		<ul style="list-style-type: none"> • <i>Analysis interwoven with existing theories and other relevant explanatory literature drawn from similar settings and studies</i> • <i>Discussion of how explanatory propositions/ Emergent theory may fit with other contexts</i> • <i>Limitations /weaknesses of study clearly outlined</i> • <i>Clearly resonates with other knowledge and experience</i> • <i>Results/ Conclusions obviously supported by evidence</i> • <i>Interpretation plausible and ‘makes sense’</i> • <i>Provides new insights and increases understanding</i> • <i>Significance for current policy and practice outlined</i> • <i>Assessment of value/ empowerment for participants</i> • <i>Outlines further directions for investigation</i> • <i>Comment on whether aims/ purposes of research were achieved</i>
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Appendix 3: Overview of 7 studies excluded from meta-ethnography

Authors (Year)	Country	Method	Quality Rating	Walsh & Downe (2006) Criteria not met	Include in Synthesis?
Case-Smith (2004)	USA	Ethnographic approach	10/12	Lack of data to support interpretation and no evidence of researcher reflexivity	No
Pitchforth et al (2011)	United Kingdom	Constant Comparative method	10/12	Design not explicit and no evidence of researcher reflexivity	No
Sullivan-Bolyai et al (2003)	USA	Naturalistic Inquiry	10/12	No clear audit trail and not enough data used to support interpretation	No
Todd, Welsh & Moriarty (2002)	United Kingdom	Descriptive Qualitative Design	10/12	Choice of design not clearly justified and no evidence of researcher reflexivity	No
Gannoni & Shute (2010)	Australia	Thematic Analysis	9/12	Design and analysis not made explicit, and no evidence of researcher reflexivity	No
Maciver, Jones & Nicol (2010)	United Kingdom	"General synthesis of techniques and analytic strategies common to applied qualitative research"	9/12	Design not apparent, analytic approach not appropriate & No evidence of researcher reflexivity	No
Maltby, Kristjanson & Coleman (2003)	USA	"Qualitative approach"	6/12	Study not thoroughly contextualised by existing literature, sampling strategy not appropriate, 2 aspects of interpretation not appropriate, no evidence of researcher reflexivity, no demonstration of ethical sensitivity	No

Parental experiences of caring for children with Cardiac Arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

ABSTRACT

The prevalence of paediatric cardiac arrhythmias is increasing, and management of some arrhythmias now involves giving the family an automatic external defibrillator (AED) to use if the child experiences a sudden cardiac arrest out of the hospital. Despite the significance of this experience for parents and the impact this may have on their children, no literature exists exploring their experience of caring for a child with cardiac arrhythmia who has an AED. The aim of this study will therefore be to explore the experiences of parents caring for a child with cardiac arrhythmia who has an AED. This will be done by conducting in-depth semi-structured interviews with six to eight main caregivers. Interpretative Phenomenological Analysis (IPA; Smith & Eatough, 2007) will be used to analyse the data. A greater understanding of parents' experiences in this context can be used to inform services about how they can best support and meet the needs of children with cardiac arrhythmia who have an AED, and their families.

INTRODUCTION

Over 700,000 people in the UK have a cardiac arrhythmia (Department of Health, 2005). Cardiac arrhythmia is an umbrella term used to describe a number of conditions where the heart muscle contracts either too slowly (called bradycardia), too quickly (called tachycardia), or irregularly because of a disturbance in the heart's normal electrical activity (Department of Health, 2005). This can be due to a genetic disorder or acquired condition (Hanash & Crosson, 2010). Consequences of arrhythmias can range in severity from minor discomfort to the risk of sudden death (Department of Health, 2005). Although many arrhythmias are benign, they may have a significant impact on a person's quality of life. Symptoms include fainting, light headedness, dizziness, sensations of heart flutters, shortness of breath, chest-pain, weakness, fatigue, and intolerance for activity (American Heart Association, 2013).

Arrhythmias in children are less common than in adults, but are increasing due to successful repair of congenital heart diseases (Doniger & Sharieff, 2006). Types of arrhythmias found in children include Supraventricular tachycardia (SVT), sinus bradycardia, Long Q-T Syndrome, Wolff-Parkinson-White Syndrome, Ventricular Tachycardia, Sick Sinus Syndrome and Complete Heart Block (American Heart Association, 2013). Treatment of these arrhythmias ranges widely depending on the cause and severity, and can include lifestyle changes, medication, a pacemaker to help the heart beat more regularly, external cardiac defibrillation, an implanted cardioverter defibrillator (ICD), cardiac ablation, and surgery (American Heart Association, 2013).

Cardiac defibrillation is necessary if the child experiences a cardiac arrest. For this reason, children who are deemed to be at highest risk of sudden death are fitted with an Implanted Cardioverter Defibrillator (ICD). Some other children with cardiac arrhythmia are also deemed to be at risk of sudden death, however not high enough to warrant an ICD. In the case of these children, their families are offered an automatic external defibrillator (AED) to take home and use if needed. Much of the practice of external defibrillation with children is derived from adult studies, and therefore there are still questions regarding the optimal techniques for paediatric defibrillation and management (Haskell & Atkins, 2010).

A significant body of quantitative research already exists in relation to the experience of living with an ICD (e.g. De Maso, Lauretti, Spieth et al., 2004; Dunbar, Dougherty, Sears et al., 2012). Additionally, a qualitative study carried out by Rahman and colleagues (Rahman, Macciocca, Sahhar et al., 2011) examined the experience of parents caring for a child with an ICD, and the experience of the children themselves of living with the ICD. However, parents of children with cardiac arrhythmia and an ICD will arguably have a different experience from those parents of children with cardiac arrhythmia and an AED. Interestingly, no literature exists, to the author's knowledge, exploring the experience of parents caring for a child with cardiac arrhythmia and an AED.

There is however a small body of literature examining the impact on adult patients and their family members of being provided with an AED in their home. The results

are mixed. One small study involving interviews with post myocardial infarction patients found that AEDs were highly valued by the patients and their partners, and increased their perceived control over their heart disease (Chen, Eisenberg & Meischke, 2002). Slightly different results were found in a randomised study by Cagle, Diehr, Meischke and colleagues (2007) comparing the quality of life in patients assigned to either cardiopulmonary resuscitation (CPR) or CPR/AED training. They found that patients in the AED group reported worse scores on quality of life, particularly in those subscales relating to social functioning, than the CPR group. Finally, a larger longitudinal observational study, comparing the long-term effects of CPR training and CPR/ AED training on anxiety and depression of patients and of their partners, found that the anxiety of partners in the CPR/AED group increased slightly over time. This was in contrast to the CPR group, in which partners' anxiety decreased significantly over 2 years of follow-up (Thomas, Friedmann & Lee et al., 2011). This led the study's authors to recommend the assessment of anxiety in partners of patients who receive AEDs, and consideration of strategies to reduce their anxiety (Thomas et al., 2011). In contrast to the study by Cagle et al. (2007), Thomas and colleagues (2011) found no evidence to suggest that home AEDs caused distress among the patients. Despite mixed results, these studies suggest that a patient's experience of being offered an AED and the caregiver's experience of being trained and expected to use it, is a significant one worthy of further exploration.

When taking the above findings into account, and when considering a parent's role in caring for a child with a chronic health condition, the importance of exploring parental experiences of caring for a child with cardiac arrhythmia who has an AED becomes apparent. Parents of children with health conditions are responsible not only for the physical care of their children, but for dealing with medical, educational, and other service providers. They are responsible for providing support to their children to help them cope with the physical and emotional demands of their illness; and for managing competing family demands.

The literature has continuously revealed a myriad of stressors that parents experience when caring for a child with chronic illness. These include financial stress, role strains within the family unit, marital separations, adjustment to working

with the medical system, interruptions in daily routines and future plans, and the general uncertainty with regard to the child's prognosis (Brown, Wiener & Kupst et al., 2008). Experiencing these potential stressors may lead directly and indirectly to anxiety, depression, post-traumatic stress, hopelessness, and feelings of loss of control (Brown et al., 2008).

In general, the literature suggests a reciprocal relationship between chronic illness and parental adaptation, whereby the child's illness impacts on parents' functioning and parental functioning impacts on child adaptation (Brown et al., 2008). This may occur even in the absence of a condition with serious implications for the child's functioning or ability. For example, increased parental perceptions of child vulnerability have been found to relate to elevated levels of social anxiety in children, even after controlling for child age and disease severity (Anthony, Gil & Schanberg, 2003).

A metasynthesis carried out by Coffey (2006) of 11 qualitative studies examining the experience of parents caring for a child with chronic illness found several common themes across all studies. According to the metasynthesis, parents shared common feelings of grief and fear around the diagnosis and management of the child's illness, and there was a clear need for support in the early stages. Exhaustion, constant worry, and carrying a burden are themes that showed up repeatedly in all the studies. Depression with suicidal ideation was also present for some parents (Coffey, 2006). Thus, it is apparent that parents can experience a great deal of distress in relation to their child's illness, and that support from services is crucial in helping them manage this.

As cardiac arrhythmia arguably differs from other chronic illnesses, and the experience of having an ICD will be different from having an AED, it would be important to explore and gain an understanding of parents' experiences of caring for a child with cardiac arrhythmia who has an AED. As mentioned previously, no literature exists, to the author's knowledge, exploring the experience of parents caring for a child with cardiac arrhythmia who has an AED. A greater understanding of parents' experiences in this context can be used to inform services about how they can best support and meet the needs of children with cardiac arrhythmia who

have an AED, and their families. As suggested in the literature, parental coping may impact upon child coping, and this is another key reason for exploring parental experiences within this population.

AIMS AND HYPOTHESES

The aim of this study will be to explore the experiences of parents caring for a child with cardiac arrhythmia who has an AED. Specifically, the study will aim to explore five key areas:

- 1) What is the parent's understanding of their child's condition?
- 2) What is the impact on the parent of having a child with cardiac arrhythmia who has an AED?
- 3) What is the parent's perception of the impact of cardiac arrhythmia and the AED on their child?
- 4) What is the parent's perception of the impact on the rest of the immediate family (e.g. other parent and children) of having a child with cardiac arrhythmia who has an AED?
- 5) What coping methods do parents use in relation to these areas, and what support might they benefit from?

PLAN OF INVESTIGATION

Participants

Between six to eight parents of children and young people in Scotland diagnosed with cardiac arrhythmia, and who have an AED, will be recruited to take part in the study. The main caregiver will be invited to take part.

Inclusion criteria

The main caregivers of children and young people with a principal diagnosis of cardiac arrhythmia, and who have an AED, will be invited to participate in the study. Main caregivers will be distinguished by asking the parents to identify who the main caregiver is. The children and young people with cardiac arrhythmia must be under 18 years old. There is no age limit for the primary caregivers.

Exclusion Criteria

Main caregivers will be excluded from the study if they do not speak English fluently. This is because the interviews will be conducted in English and unfortunately there is no budget available for employment of interpreters. Main caregivers will be excluded from the study if their child does not have an AED.

Recruitment Procedures

Dr Karen McLeod, Consultant Cardiologist, and Sister Eileen Fern, Clinical Nurse Specialist, will send out the invitation letter and information sheet about the current study by post to all parents of children routinely attending the RHSC who meet the inclusion criteria. The information sheet will explain what the current study involves. It will ask the main caregiver of the child with cardiac arrhythmia to either phone Sister Eileen Fern from the cardiac team, or to complete and return a response form in a pre-addressed and postage paid envelope, to express their interest in taking part in the study. Sister Eileen Fern will then pass their details on to the researcher who will contact them by telephone approximately a week later to provide further information about the study and answer any questions. The information sheet and the researcher will emphasise that participation is voluntary and that they may choose to withdraw at any time. The information sheet will also explain that, due to study design, a maximum of eight caregivers will be interviewed, and therefore recruitment will be on a first-come, first-served basis.

If the caregiver is still interested in participating after being followed up by the researcher and having their questions responded to, an appointment will be arranged to carry out the interview at RHSC during office hours. If it is not possible for a participant to attend the RHSC during office hours, but they are still keen to engage in the research study, a telephone interview will be arranged. It is recognised that a telephone interview may make it more difficult for the interviewer to establish rapport with the participant, and will prevent the interviewer from observing the participant's non-verbal communication which can provide a context for subsequent analysis (Turner, Barlow & Ilbery, 2002). However, it is felt that conducting a telephone interview is a superior option to denying participants the chance to discuss their experiences if they are keen to engage and cannot attend in person due to circumstances out with their control. Previous studies employing IPA have used telephone interviews with success, and researchers describe having

established strong relationships with the participants over the telephone and demonstrate generating rich and meaningful data from the interviews (e.g. Jenkins & Ogden, 2011; Ogden & Hills, 2008; Turner et al., 2002).

Written consent will be obtained from participants prior to the interview. In the case of a telephone interview, written consent will be obtained via post from participants prior to the interview taking place, using a postage-paid envelope. If more than eight caregivers register interest, the extra caregivers will be informed via telephone or letter that eight others have already replied, and that they will be contacted again if any of these eight choose to withdraw from the study.

Measures

An interview schedule to guide the semi-structured interviews will be devised from the existing literature on experiences of parents of children with a chronic illness. The interview schedule will be used flexibly. The role of the interviewer as an active listener may lead to the interviewer moving away from the interview schedule in order to follow other unanticipated issues raised by the participant (Smith, Flowers & Larkin, 2009). The aim of the interview is to explore what the participant feels is important about their experiences in relation to caring for a child with cardiac arrhythmia and an AED, and therefore the interview schedule will not constrict this exploration.

Design

This study will employ Interpretative Phenomenological Analysis (IPA). IPA aims to explore an important aspect of the research participant's life, for example their experience of an illness, from their personal perspective, whilst recognising that how this account is interpreted by the researcher(s) is inevitably influenced by their own cognitions and past experiences, plus the process of interaction between the researcher(s) and the participant (Smith & Eatough, 2007).

Research procedures

The researcher will conduct interviews of approximately one hour with each participant, using the developed interview schedule as a guide. Interviews will be recorded on a digital voice recorder and then transcribed verbatim by the researcher,

with all identifiers of person and place removed. The transcripts will then be analysed using IPA (Smith & Eatough, 2007).

Data Analysis

Data analysis will be conducted in accordance with the IPA stages outlined by Smith et al (2009). At least two transcripts will be independently analysed by both the researcher and the project supervisors, and themes resulting from the independent analyses will be compared to check the reliability of the primary analyst.

Justification of Sample Size

IPA is an idiographic approach, concerned with understanding particular phenomena in specific contexts. Therefore small sample sizes are recommended to allow for detailed case-by-case analysis, to form an in-depth and rich understanding of the perceptions and experiences of the participants involved (Smith et al., 2009). Analysis using IPA is described as potentially being a “difficult, creative, intense and conceptually demanding” experience (Smith et al., 2009, pg.80) and an iterative, time-consuming process (Smith et al., 2009). It is therefore felt that recruiting between six and eight participants would be optimal for the purpose of this study, as this will enable the researcher to gain an in-depth understanding, and carry out a rich and detailed analysis, of the experiences of the participants. According to cardiac medical staff at the RHSC there are only 17 families in Scotland who have a child with a cardiac arrhythmia and an AED, and therefore recruiting 8 participants would provide data from approximately half of the available sample.

Settings and Equipment

Invitation letters, Information forms, response forms and consent forms will be prepared for the purpose of the study. Interviews will be conducted within the Clinical Psychology Department clinic rooms at the RHSC, Yorkhill Hospital or the Department of Child & Family Psychiatry/Caledonia House, Yorkhill Hospital. Full permission to use these clinic rooms has been granted by the Clinical Psychology team lead. If a participant is unable to attend Yorkhill Hospital to engage in the interview, a telephone interview will be arranged. All telephone interviews will be carried out from a Clinical Psychology clinic room within Yorkhill Hospital, and therefore the cost of the telephone call will be subsumed within the overall running

costs of the Clinical Psychology Department. The Clinical Psychology Department is supportive of this and has given permission for telephone calls to be carried out, given the beneficial nature of the research to the work of the department. Recording and transcribing equipment will be required.

HEALTH AND SAFETY ISSUES

Researcher Safety Issues

No safety issues are anticipated; however the interviews will be conducted within a clinic room at Yorkhill Hospital, with available practitioners nearby should any issues occur.

Participant Safety Issues

No participant safety issues are anticipated. The interviewer is a Trainee Clinical Psychologist, and therefore is skilled at undertaking interviews sensitively, and picking up on any signs of distress or upset. Participants will be informed that they may stop the interview at any time, and if the interviewer identifies any signs of distress during the interview, the interview will be stopped and further support will be offered to the participant via the Clinical Psychology department at RHSC or through adult mental health services, the arrangement of which RHSC Clinical Psychology will facilitate. Data will be anonymised following transcription and stored on a password protected, encrypted computer.

ETHICAL ISSUES

The study has been verbally approved by the Paediatric Cardiac Team at RHSC, Yorkhill Hospital. Researcher and Participant safety will be carefully considered and managed according to local protocols. Ethical approval will be sought from Integrated Research Application System (IRAS).

FINANCIAL ISSUES

Recording and transcribing equipment can be borrowed from the Mental Health and Wellbeing Research department, University of Glasgow. White paper, printing, envelopes and postage for participant letters and information packs are the only anticipated costs. The total cost is estimated to be about £22.76.

TIMETABLE

The MRP proposal will be submitted in April 2013. Application to IRAS will take place in October 2013. This should allow recruitment to begin in November 2013. Data collection will be completed by February 2014 allowing for submission in July 2014.

PRACTICAL APPLICATIONS

This research will provide an insight into parents' experiences of caring for a child with cardiac arrhythmia who has an AED, which will be disseminated to practitioners working with this population. A greater understanding of parents' experiences can be used to inform services about how they can best support and meet the needs of children with cardiac arrhythmia and their families. A Paediatric Clinical Psychology service is in the process of being established within the RHSC cardiology department, and this research will help directly inform the new service development.

REFERENCES

- American Heart Association (2013). *What is an Arrhythmia?* [Online] Available at: <http://www.heart.org/HEARTORG/Conditions/Arrhythmia> [Accessed 21 January 2013].
- Anthony, K., Gill, K. & Schanberg, L. (2003). Brief Report: Parental Perceptions of Child Vulnerability in Children with Chronic Illness. *Journal of Pediatric Psychology*, 28 (3), 185-190.
- Brown, R., Wiener, L. Kupst, M., Brennan, T., Behrman, R., Compas, B., Elkin, D., Fairclough, D., Friebert, S., Katz, E., Kazak, A., Madan-Swain, A., Mansfield, N., Mullins, L., Noll, R., Patenaude, F., Phipps, S., Sahler, O., Sourkes, B. & Zeltzer, L. (2008). Single Parenting and Children with Chronic Illness: An Understudied Phenomenon. *Journal of Pediatric Psychology*, 33(4), 408–421.
- Cagle, A.J., Diehr, P., Meischke, H., Rea, T., Olsen, J., Rodrigues, D., Yakovlevitch, M., Amidon, T. & Eisenberg, M. (2007). Psychological and social impacts of automated external defibrillators (AEDs) in the home. *Resuscitation*, 74, 432-438.
- Chen, M.A., Eisenberg, M.S. & Meischke, H. (2002). Impact of in-home defibrillators on postmyocardial infarction patients and their significant others; an interview study. *Heart and Lung*, 31(3), 173-185.
- Coffey, J.S. (2006). Parenting a child with chronic illness: a metasynthesis. *Journal of Paediatric Nursing*, 32 (1), 51-59.
- DeMaso, R., Lauretti, A., Spieth, L., Van Der Feen, J., Jay, K., Gauvreau, K., Walsh, E. & Berul, C. (2004). Psychosocial factors and quality of life in children and adolescents with implantable cardioverter-defibrillators. *The American Journal of Cardiology*, 93, 582- 587.
- Department of Health (2005) *National Services Framework for Coronary Heart Disease. Chapter Eight: Arrhythmias and Sudden Cardiac Death*. [Online] Available at: <http://www.c-r-y.org.uk/NSF%20Chapter%208.pdf> [Accessed 1 December 2012].
- Doniger, S.J. & Sharieff, G.Q. (2006). Pediatric dysarrhythmias. *Pediatric Clinics of North America*, 53, 85-105.

Dunbar, S., Dougherty, C., Sears, S., Carroll, D., Goldstein, N., Mark, D., McDaniel, G., Pressler, S., Schron, E., Wang, P. & Zeigler, V. (2012). Educational and Psychological Interventions to improve outcomes for recipients of implantable cardioverter defibrillators and their families. A scientific statement from the American heart association. *Circulation*, 126, 2146-2172.

Jenkins, J & Ogden, J. (2011). Becoming whole again: A qualitative study of women's views of recovering from anorexia nervosa. *European Eating Disorders Review*. John Wiley & Sons Ltd & Eating Disorders Association.

Hanash, C.R. & Crosson, J.E. (2010). Emergency diagnosis and management of pediatric arrhythmias. *Journal of Emergency, Trauma and Shock*, 3 (3), 251-260.

Haskell, S. & Atkins, D. (2010). Defibrillation in children. *Journal of Emergency, Trauma and Shock*, 3(3), 261-266.

Ogden, J., & Hills, L. (2008). Understanding sustained changes in behaviour: The role of life events and the process of reinvention. *Health: An International Journal*, 12, 419–437.

Rahman, B., Macciocca, I., Sahhar, M., Kamberi, S., Connell, V. & Duncan, R. (2011). Adolescents with Implantable Cardioverter Defibrillators: A Patient and Parent Perspective. *Pacing and Clinical Electrophysiology*, 35, 62-72.

Smith, J. A., & Eatough, V. (2007). Interpretative Phenomenological Analysis. In Lyons, E., & Coyle, A., (Eds.) *Analysing Qualitative Data in Psychology* (Chapter 3). London: Sage Publications.

Smith, J.A., Flowers, P. & Larkin, M. (2009). *Interpretative Phenomenological Analysis: Theory, method and research*. London: Sage Publications.

Thomas, S., Friedmann, E., Lee, H-J., Son, H. & Morton, P. (2011). Changes in anxiety and depression over 2 years in medically stable patients after myocardial infarction and their spouses in the Home Automatic External Defibrillator Trial (HAT): a longitudinal observational study. *Heart*, 97, 371-381.

Turner, A, Barlow, J. & Ilbery, B. (2002). Play hurt, live hurt: living with and managing osteoarthritis from the perspective of ex-professional footballers. *Journal of Health Psychology*, 7, 285-301.

Appendix 5: Guidelines for Submission to The Journal of Pediatric Psychology

Instructions to Authors

The *Journal of Pediatric Psychology* is an official publication of the Society of Pediatric Psychology, Division 54 of the American Psychological Association. JPP publishes articles related to theory, research, and professional practice in pediatric psychology.

Types of Manuscripts:

- Original research, including case studies
- Review articles
- Commentaries

Manuscript preparation: General Instructions

Full instructions for uploading data and files etc. are given on Manuscript Central at the website under Instructions for online submission:

http://www.oxfordjournals.org/our_journals/jpepsy/for_authors/submission_online.html

Organization of manuscripts

Manuscript Central will guide authors through the submission steps, including: Abstract, Keyword selection, and the Manuscript. The manuscript must contain an Introduction, Methods, Results, Discussion, Acknowledgements and Reference List.

Length of manuscript: Original research articles should not exceed 25 pages, in total, including title page, references, figures, tables, etc. In the case of papers that report on multiple studies or those with methodologies that necessitate detailed explanation, the authors should justify longer manuscript length to the Editor in the cover letter. Case reports should not exceed 20 pages. Review articles should not exceed 30 pages. Commentaries should not exceed 4 pages. The Journal of Pediatric Psychology no longer accepts brief reports but will accept manuscripts that are shorter in length than the 25 page manuscripts.

Manuscripts (text, references, tables, figures, etc.) should be prepared in detailed accord with the Publication Manual of the American Psychological Association (6th ed.). There are two exceptions:

(a) The academic degrees of authors should be placed on the title page following their names, and

(b) a structured abstract of not more than 150 words should be included. The abstract should include the following parts:

- (1) Objective (brief statement of the purpose of the study);
- (2) Methods (summary of the participants, design, measures, procedure);
- (3) Results (the primary findings of this work); and
- (4) Conclusions (statement of implications of these data).

Key words should be included, consistent with APA style. Submissions should be double-spaced throughout, with margins of at least 1 inch and font size of 12 points (or 26 lines per page, 12-15 characters per inch). Authors should remove all identifying information from the body of the manuscript so that peer reviewers will be unable to recognize the authors and their affiliations. E-mail addresses, whenever possible, should be included in the author note.

Informed consent and ethical treatment of study participants. Authors should indicate in the Method section of relevant manuscripts how informed consent was obtained and report the approval of the study by the appropriate Institutional Review Board(s). Authors will also be asked to sign a statement, provided by the Editor that they have complied with the American Psychological Association Ethical Principles with regard to the treatment of their sample.

Clinical relevance of the research should be incorporated into the manuscripts. There is no special section on clinical implications, but authors should integrate implications for practice, as appropriate, into papers.

Terminology should be sensitive to the individual who has a disease or disability. The Editors endorse the concept of "people first, not their disability." Terminology should reflect the "person with a disability" (e.g., children with diabetes, persons with HIV infection, families of children with cancer) rather than the condition as an adjective (e.g., diabetic children, HIV patients, cancer families). Nonsexist language should be used.

Appendix 6: University of Glasgow Approval Letter



University of Glasgow | College of Medical,
Veterinary & Life Sciences

TMcM/LC

5th August 2013

Sonia Anker-Petersen
Flat 0/2
60 Fergus Drive
Glasgow
G20 6AW

Dear Sonia,

Doctorate in Clinical Psychology Major Research Project
Parental experiences of caring for children with Cardiac Arrhythmia who have an Automatic External
Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

The above project has been reviewed by your University Research supervisor and by a member of staff not involved in your project and has now been deemed fit to proceed to ethics.

Congratulations and good luck with the study.

Yours sincerely,

T M McMillan
Professor of Clinical Neuropsychology
Research Director

Doctorate in Clinical Psychology
Programme Director: Dr Hamish McLeod

Mental Health and Wellbeing
Admin Building, Gartnavel Royal Hospital
1055 Great Western Road
GLASGOW G12 0XH
Direct line: +44(0) 141 211 3920/0607 Fax: +44(0) 141 211 0356
Email: mhwb-clinpsy-students@glas.ac.uk

The University of Glasgow, charity number SC004401

Appendix 7: Nottingham REC Approval letter



Health Research Authority

NRES Committee East Midlands - Nottingham 2

The Old Chapel
Royal Standard Place
Nottingham
NG1 6FS

Telephone: 0115 8839697

31 December 2013

Miss Sonia Anker-Petersen
Mental Health & Wellbeing
Gartnavel Royal Hospital, Admin. Building
Glasgow
G12 0XH

Dear Miss Anker-Petersen,

Study title:	Parental experiences of caring for children with Cardiac Arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis
REC reference:	13/EM/0448
Protocol number:	n/a
IRAS project ID:	137905

Thank you for your letter of 20th December 2013. I can confirm the REC has received the documents listed below and that these comply with the approval conditions detailed in our letter dated 26 November 2013.

Documents received

The documents received were as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter		20 December 2013
Letter of invitation to participant	3	20 December 2013

Approved documents

The final list of approved documentation for the study is therefore as follows:

<i>Document</i>	<i>Version</i>	<i>Date</i>
Covering Letter		20 December 2013
Interview Schedules/Topic Guides	3	31 October 2013
Investigator CV	(Sonia Anker-Petersen)	14 November 2013
Investigator CV	(Dr Sarah L Wilson)	09 August 2007

Investigator CV	(Dr Kathleen A McHugh)	19 September 2013
Letter of invitation to participant	3	20 December 2013
Other: Participant Response Form	3	21 October 2013
Participant Consent Form	4	07 November 2013
Participant Information Sheet	4	21 October 2013
Protocol	3	18 October 2013
REC application		14 November 2013

You should ensure that the sponsor has a copy of the final documentation for the study. It is the sponsor's responsibility to ensure that the documentation is made available to R&D offices at all participating sites.

13/EM/0448	Please quote this number on all correspondence
------------	--

Yours sincerely,



Rachel Nelson
REC Assistant

E-mail: NRESCCommittee.EastMidlands-Nottingham2@nhs.net

Copy to: Miss Sonia Anker-Petersen

Ms Joanne McGarry

Appendix 8: GG&C R&D Approval Letter



Coordinator/Administrator: JMcG/ LR
Direct Line: 0141 211 8548
E-mail: Joanne.McGarry@ggc.scot.nhs.uk
Website: www.nhsggc.org.uk/r&d

R&D Management Office
Western Infirmary
Tennent Institute
1st Floor, 38 Church St
Glasgow
G11 6NT

21st Feb 2014

Dr Kathleen McHugh
Clinical Psychologist
Dept of Clinical Psychology
Royal Hospital for Sick Children
Dalnair Street
Glasgow
G3 8SJ

NHS GG&C Board Approval

Dear Dr McHugh

Study Title: Parental experiences of caring for children with Cardiac Arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

Chief Investigator: Miss Sonia Anker-Peterson

GG&C HB site: RHSC

Sponsor: NHS GG&C Health Board

R&D Reference: GN13CP380

REC Ref: 13/EM/0448

Protocol no: V3 dated 18/10/13

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 related solely 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.nhsggc.org.uk/content/default.asp?page=s1411), evidence of such training to be filed in the site file.

2. **For all studies** the following information is required during their lifespan.
 - a. Recruitment Numbers on a monthly basis
 - b. Any change of staff named on the original SSI form
 - c. Any amendments – Substantial or Non Substantial
 - d. Notification of Trial/study end including final recruitment figures
 - e. 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



**Joanne McGarry
Research Co-ordinator**

**CC: Miss Sonia Anker-Peterson, Chief Investigator/Student, Glasgow
Dr Sarah Wilson, Academic Supervisor, Glasgow**

Appendix 9: Participant Invitation Letter

Experience of Caring for children with cardiac arrhythmia and an AED1: Letter of Invitation,
version 3. Date: 20.12.13



Cardiology Department
RHSC
Yorkhill
Glasgow, G3 8SJ

Date:

Dear *(insert family name)*

I would like to invite you to take part in a research study being undertaken by Sonia Anker-Petersen, a Trainee Clinical Psychologist with NHS Greater Glasgow and Clyde and the University of Glasgow. She is undertaking a research project in partnership with the Paediatric Cardiac Team at Royal Hospital for Sick Children (RHSC), Yorkhill Hospital, as part of her Doctorate in Clinical Psychology. The aim of the study is to gain a better understanding of parental experiences of caring for a child with cardiac arrhythmia who has an automatic external defibrillator. We are looking at how living with a cardiac arrhythmia and an automatic external defibrillator might affect feelings, behaviour and coping in children, young people and their families.

Because your child is diagnosed with a cardiac arrhythmia and has an automatic external defibrillator, you are being invited to take part in this study. Before you decide, it is important for you to understand what we are asking you to do and why. Please take time to read the enclosed information sheet carefully and discuss it with other people, including your child, if you wish. If anything is not clear or you want more information please do not hesitate to contact Sonia. You can also choose to contact Dr Kenneth Mullen, Associate Academic at the Institute of Health and Wellbeing, University of Glasgow School of Medicine. Dr Mullen is an independent contact for the study as he is not involved in the research, but is aware of what it involves. Contact details for both of them can be found in the enclosed Participant Information Sheet.

Asking you to take part in this study does not mean we think you or your child are not coping with cardiac illness. We are asking everyone who has a child with cardiac arrhythmia and an automatic external defibrillator who attends RHSC to take part. You can say no if you don't want to take part. This will not affect your child's clinical care in any way.

Thank you for reading this.

Experience of Caring for children with cardiac arrhythmia and an AED1: Letter of Invitation,
version 3. Date: 20.12.13

Yours sincerely

Dr Karen MacLeod
Consultant Cardiologist
RHSC, Yorkhill Hospital

Appendix 10: Participant Information Sheet

Experience of Caring for children with cardiac arrhythmia and an AED1: Participant Information Sheet, version 4. Date: 21.10.13



INFORMATION SHEET FOR CAREGIVERS

Study Title:

Parental experiences of caring for children with cardiac arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

I would be grateful if you could read over this information leaflet. Reading through it will probably take about 10 minutes. My name is Sonia Anker-Petersen and I am a Trainee Clinical Psychologist with NHS Greater Glasgow and Clyde and the University of Glasgow. I am undertaking a research project in partnership with the Paediatric Cardiac Team at Royal Hospital for Sick Children (RHSC), Yorkhill Hospital, as part of my Doctorate in Clinical Psychology.

I would like to invite you to take part in our research study. All main caregivers of a child with cardiac arrhythmia who has an automatic external defibrillator (AED), and who attends RHSC, are being invited to take part in the study. Before you decide if you would like to take part, we would like you to understand why the research is being done and what it would involve for you. Please take time to read the following information carefully and discuss it with other people if you wish. If anything is not clear or you want more information please do not hesitate to contact me. I would be happy to go through the information sheet with you and answer any questions you may have. Thank you for reading this.

What is the study about?

The aim of the study is to gain a better understanding of parents' experiences of caring for a child with cardiac arrhythmia who has been given an AED. I would also like to explore what you think might be helpful in terms of supporting children with cardiac arrhythmia, and their families, in the future. The findings from the study will help inform the development and practice of the new Clinical Psychology Service currently being established within the RHSC cardiology department.

Why am I being asked to take part?

You are being asked to take part because you have a child diagnosed with cardiac arrhythmia who has an AED, and you have identified yourself as the main caregiver of that child. We would hope to interview up to 8 main caregivers as part of this study.

Do I have to take part?

No, you do not have to take part in the study. Participation is completely voluntary. It is up to you to decide whether or not you join the study. We will describe the study and go through the information sheet. If you decide to take part in the study, you will

be asked to sign a consent form. If you agree to take part, you can withdraw from the study at any time up until I start writing up the final report, which will commence in May 2014. You do not have to give a reason for changing your mind. It will not affect you or your child's medical care or treatment in any way if you say no to the study.

What would I have to do if I take part in the study?

You would be asked to take part in an interview with me within a clinic room in the RHSC, Yorkhill Hospital, which is expected to last for about 1 hour. If you are unable to attend the RHSC, a telephone interview can be arranged. The interview will be digitally recorded. The interview will focus on getting an understanding of your experiences as a caregiver for your child, what impact you feel your child's cardiac arrhythmia and AED has had on you and your family, and what support might be helpful to your family and other families in a similar situation.

Who would know I was taking part?

The Cardiac Team at RHSC, Yorkhill Hospital would know that you were considering taking part in the study, because they would only be allowed to pass me your contact details if you have expressed interest in taking part and given consent to them passing me your details. However we will follow ethical and legal practice to ensure that all information about you will be handled in confidence. The information that you provide in the interview will be made anonymous so that no one can identify what you specifically have said. I will need to know your name and date of birth, and your child's age, for demographic reasons and for follow up as required. However these will not be shared with others.

I would only have to break anonymity if I became concerned that you or someone else was at risk of harm. In these circumstances, I would need to share my concerns but I would tell you before I did this. In the first instance, I would share my concerns with the senior clinical psychologist attached to the study and it may be that we feel it necessary to contact other agencies, for example, your GP or social work.

What will happen to the information I provide?

The interview will be digitally recorded. The recordings will be transcribed (typed up) and made anonymous by me. The recordings will then be destroyed. The anonymous transcripts will be stored on an encrypted password protected computer. Only my research supervisors and I will have access to the anonymous transcripts. The anonymous information will be analysed and presented, along with information from other caregiver interviews, in the form of a report. This report will be submitted to the University of Glasgow in part fulfilment of the Doctorate in Clinical Psychology and for publication in a scientific journal. A presentation summarising the report will also be provided to the Paediatric Cardiac Team at RHSC. Within the report, anonymous quotes of what you have said may be used. Neither your name nor your child's name will appear in any reports or publications arising from the research. You will be provided with a summary of the results via post if you wish, and access to the full report will be available via the Glasgow University Library.

Are there any benefits to taking part?

We cannot promise the study will help you directly but the findings from the study will help people involved in your child's care, and other children's care, to understand

the experiences of caregivers and families of having a child with cardiac arrhythmia and an automatic external defibrillator. The study will help inform the development and practice of the new Clinical Psychology Service currently being established within the RHSC cardiology department, which can provide important support to families with a child with cardiac arrhythmia. Additionally, if this study is published in a scientific journal, it will contribute to the wider research literature and could contribute to wider developments in the psychological care of cardiac arrhythmia patients and their families.

Are there any disadvantages or risks to taking part?

Although no risks are anticipated, it is possible that the interview may trigger upsetting thoughts or feelings that may be difficult for you to talk about. If this is the case, and you wish to stop, you can end the interview at any time. If you need a break during the interview this is fine. If you wish to be referred to a Clinical Psychologist for further support regarding the issues discussed, this will be arranged.

Will I be paid for taking part?

No, as this is a voluntary study we cannot offer any payment. Unfortunately we cannot provide travel expenses to attend the hospital for interview. However we will try our best to arrange the interview at a time that suits you and, if possible, when you are attending for your child's routine appointment anyway. Alternatively, a telephone interview can be arranged.

Who has reviewed the study?

All research in the NHS is looked at by an independent group of people, called a Research Ethics Committee, to protect your interests. This study has been reviewed and given favourable opinion by the Nottingham Research Ethics Committee. The study has also been approved by the University of Glasgow, and NHS Greater Glasgow and Clyde Research and Development.

Independent Contact for further information

Dr Kenneth Mullen, Associate Academic at the Institute of Health and Wellbeing, is not involved with this study but is aware of what it involves and would be happy to be contacted as an independent person who can discuss what the study involves if you are undecided about taking part. You can call Dr Mullen on 0141 211 3932 or email him at Kenneth.Mullen@glasgow.ac.uk.

What do I do now?

If you are interested in taking part in the study, please complete the attached form and return it in the enclosed envelope. Alternatively, you can phone Sister Eileen Fern, Clinical Nurse Specialist within the Cardiac Team on 0141 _____ and leave a message with your contact details expressing your interest in the study. I will then contact you by telephone to answer any other questions that you may have about the study and, if you are still interested in taking part following this, we can arrange an appointment for the interview. When we meet, I will go over the information sheet with you again and ask you to sign a consent form to show that you have read and understood the information provided to you and that you agree to take part in the study. If a telephone interview is arranged, I will discuss the information sheet with you on the telephone and send you a consent form in the post or via email that you will need to sign and return to me before the telephone interview.

Due to study design, a maximum of eight caregivers will be interviewed, and therefore recruitment will be on a first-come, first-served basis. If eight other caregivers have already registered interest when we receive your details, I will contact you to let you know. I will invite you to join a waiting list in case anyone else chooses to drop out of the research study.

Thank you for taking the time to read this Participant Information Sheet and for any further input you may wish to have.

Sonia Anker-Petersen
Trainee Clinical Psychologist

Contact information:
Miss Sonia Anker-Petersen
Trainee Clinical Psychologist
Mental Health & Wellbeing
Administration Building
Gartnavel Royal Hospital
1055 Great Western Road
G12 0XH

Email: s.anker-petersen.1@research.gla.ac.uk

Tel : 0141 201 0644

Appendix 11: Participant Response Form

Experience of Caring for children with cardiac arrhythmia and an AED1: Participant Response Form, version 3. Date: 21.10.13



Participant Response Form

To be completed and returned if you are interested in taking part in the study titled *"Parental experiences of caring for children with cardiac arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis"*

Name:

Address (including postcode):

Telephone Number:

Email address (optional):

Please tick the box to confirm that your child attends RHSC, is diagnosed with cardiac arrhythmia, and has been given an automatic external defibrillator related to this.

☐

Please tick the box to confirm that you regard yourself as the main caregiver for your child.

☐

Please tick the box to confirm that you are happy for Sonia Anker-Petersen, the main researcher for this study, to contact you via telephone to discuss the study further and answer any questions you may have.

☐

Are there any days and times that suit best for Sonia to phone you? Please note them below.

Demographic Details

It would be useful to gather a little more information about you and your child if you are going to take part in the study. This is to gain a better picture of who is participating in the study. All information you provide here will be made anonymous, however it is your choice whether or not you provide this information. Please take some time to read the questions below and complete the answers if you feel comfortable doing so.

Information about you:

What is your date of birth? _____

Are you: Male ☐ Female ☐

Are you: Single ☐ Cohabiting with a partner ☐ Married ☐

Divorced ☐ In a relationship ☐

What is your highest level of education?

Secondary school ☐ College ☐ Undergraduate ☐

Postgraduate ☐

Information about your child:

How old is your child? _____

Is your child: Male ☐ Female ☐

What is your child's medical diagnosis?

Appendix 12: Participant Consent Form

Experience of caring for children with Cardiac Arrhythmia and an AED1: Consent Form, version 4.
Date: 7.11.13



Parental experiences of caring for children with cardiac arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

Study Consent Form

Please initial each box if you agree:

I confirm that I have read the information sheet dated 21.10.13 (version 4) for the above study.

☐

I understand that my participation is voluntary and that I am free to withdraw at any time up until the final write up of the report, without giving a reason, and without my or my child's medical care or legal rights being affected.

☐

I confirm that I have had the opportunity to ask questions about the study and I have received satisfactory answers to all of my questions.

☐

I have received enough information about the study.

☐

Who from the research team have you spoken to about this study?

-PLEASE PRINT HIS/HER NAME

I agree to take part in this study.

☐

Name of parent/guardian

Date

Signature

Name of researcher

Date

Signature

IMPORTANT:

- One signed original to be given to participant (with a copy of the Participant Information Sheet)
- One signed original to be kept on file by the researcher

Appendix 13: Interview Schedule

Experience of caring for children with Cardiac Arrhythmia and an AED1: Questionnaire, version 3.

Date: 31.10.13



Parental experiences of caring for children with Cardiac Arrhythmia who have an Automatic External Defibrillator: An exploratory study using Interpretative Phenomenological Analysis

Interview Schedule

- ❖ **What was it like for you and your family when you learned of *(child's name)*'s condition?**
- ❖ **Possible Prompts (if needed):**
 - How did you find out about *(child's name)*'s diagnosis?
 - What was your reaction when you found out?
 - What was *(child's name)*'s reaction?
 - What did it feel like for you?
 - *For any of the above: Can you tell me more about that?*
- ❖ **What was it like for you when you were told about the AED?**
- ❖ **Possible Prompts (if needed):**
 - How do you feel about maybe having to use the AED?
 - How do you feel about the support you have received in relation to this?
 - *For any of the above: Can you tell me more about that?*
- ❖ **What has changed for you since *(child's name)* was given his/ her diagnosis and AED?**
- ❖ **Possible Prompts (if needed):**
 - Day to day life?
 - Being a parent?
 - Relationships with family?
 - Friends?
 - Work?

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- *For any of the above: Can you tell me more about that? OR How does that make you feel?*

❖ **What do you think has changed for (child's name) since getting his/her diagnosis?**

❖ Possible Prompts (if needed):

- Day to day life?
- School and other activities?
- Relationships with family and friends?
- Sense of self and self-esteem?
- Attitude and future goals?
- *For any of the above: Can you tell me more about that?
How does that make you feel?*

❖ **What do you think has changed for (child's name) since getting his/her AED?**

❖ Possible Prompts (if needed):

- Day to day life?
- School and other activities?
- Relationships with family and friends?
- Sense of self and self-esteem?
- Attitude and future goals?
- *For any of the above: Can you tell me more about that?
How does that make you feel?*

❖ **What has changed for others in your family?**

❖ Possible Prompts (if needed):

- Family's functioning?
- Relationships?
- Siblings?
- Partner?
- *For any of the above: Can you tell me more about that?
How does that make you feel?*

❖ **How have you and your family coped with this?**

❖ Possible Prompts (if needed):

- Practically?
- Emotionally?

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- Financially?
- Is there anything you wish you had done differently?
- For any of the above: *Would you mind telling me more about that?*
How does that make you feel?

❖ **What support have you found helpful?**

❖ Possible Prompt:

- *Would you mind telling me more about that?*
- *How does that make you feel?*

❖ **What support have you found unhelpful?**

❖ Possible Prompt:

- *Would you mind telling me more about that?*
- *How does that make you feel?*

❖ **We are coming to the end of the interview time now. Is there anything I haven't asked you about in relation to (child's name)'s condition and your experience, that you would like to mention?**

Appendix 14: Overview of the Omitted Themes

Impact of the genetic link

Both Mary's husband and Jane had learned that their child had developed their condition due to a genetic vulnerability that they had passed on. This appeared to have a significant impact on them and reportedly led to initial feelings of shock, guilt and devastation. It was initially very difficult for the caregivers to accept that they had passed the genetic vulnerability onto their child; they blamed themselves for their child's difficult experiences related to the condition. These feelings disappeared with time for Jane, however Mary reported that her husband still struggled with this.

Jack and Mary reported having a different perspective on learning of the genetic link, namely feeling grateful for having found out and feeling reassured at learning that no other family members were suffering from the same condition. Additionally, Jack reported that having an explanation for his child's condition helped him adjust to it.

Terri did not learn of the genetic link until after the death of her daughter-in-law, which tragically occurred due to a cardiac arrest that was caused by her undiagnosed cardiac arrhythmia. It was clear that this experience had deeply affected the whole family and therefore the genetic link had a very strong significance for Terri and her grandchildren.

Lisa and Grant described being in a slightly different position regarding genetics: they had been informed that the cause of their son's condition was due to a genetic mutation of which little is known about yet. This lack of understanding regarding the genetic mutation meant that their son was not responding as expected to medication that usually worked to treat other people with cardiac arrhythmia, leading to complications in treatment. These complications with medication were a great cause of anxiety and worry for Lisa and Grant, and it appeared that the lack of understanding about the gene fault led them to feel considerable mistrust and doubt regarding the medical professionals' ability to provide adequate medical support to their son.

Lastly, Jennifer and her husband had been through genetic testing and had been informed that there was no genetic link in their case, and therefore there was no explanation for why their son had developed his condition. Jennifer reported that not knowing the cause of her son's condition made it more difficult for her to adjust to it, and she would have preferred there to be a genetic cause so that she had some answers in relation to how it had developed.

Coping Strategies

Aside from the coping strategies discussed in the final report, caregivers reported using several other strategies. These varied between families, but included: seeking information and gathering knowledge about their child's condition, utilising support from peers and family, maintaining vigilance for any signs or symptoms of a seizure, rationalisation, trying to maintain a sense of normality for their child, being trained in CPR, sharing information with school, keeping 'on top of everything', encouraging independence in their child's management of the condition, utilising humour, looking for positives, and 'just getting on with it'. Interestingly, Terri reported not being aware of utilising any coping strategies, and stated that she just "had to cope". Despite this, it appeared that she used several of the above strategies without realising it herself.

Appendix 15: Implications for Clinical Practice

In Relation to Cardiac Arrhythmia:

- Families should be offered mental health follow-up support following diagnosis.
- Close follow-up of adolescent patients after diagnosis is particularly important given their vulnerability at this developmental stage.
- Providing psycho-education to caregivers regarding the impact of chronic illness on the child and family's development at the point of diagnosis would help them prepare for and manage any difficulties encountered.
- It is important that medical staff take measures to reduce parental uncertainty. This can be done through providing clear, basic information to caregivers when possible, discussing and encouraging problem-solving skills with caregivers, and increasing open communication with medical staff.
- Supporting caregivers to balance their child's increasing desire for autonomy with the management of their condition would be important.
- If resources allow, providing caregivers with a stethoscope and showing them how to use it may be a helpful and relatively simple way of helping caregivers to feel empowered and more in control of monitoring their child's condition.
- Ensure communication is sensitive and considerate of the child and family's needs at all times. Ensure medical staff feel confident in their communication skills and encourage extra staff training in communicating difficult news if this is indicated.
- Ensure the child and caregivers are supported in their transition from child to adult services

In Relation to the AED:

- A home visit to the family after initial training would be helpful to assess their adaptation to the device and provide appropriate follow-up support as required.
- It is recommended that an assessment of caregivers' anxiety levels regarding having to use the AED is undertaken with all families given an AED. Appropriate follow-up support with anxiety management strategies should be offered if required.

- It is recommended that annual refresher training in how to use the AED is offered to all caregivers.
- Better education and training of school staff involved in the patient's care regarding the risks of cardiac arrhythmia and the function of the AED is indicated.
- Hospital staff should undertake on-going monitoring of caregivers' perceptions of their child's condition and their understanding of the requirement of the AED to assess for any potential misunderstandings or risks related to this.
- Hospital staff should monitor the situation with school and the AED, and support caregivers in communications with school if this is necessary.
- Ensure appropriate information and preparation is given to caregivers and the child regarding the recall of the AED at the point of transition.

Appendix 16: Excerpt from Interview 2 with Lisa and Grant (Pseudonyms), pages 40-43.

Emergent Themes	Original Transcript	Exploratory comments
Inconsistency in treatment advice	L: maybe it's their interpretation. Maybe one consultant to the other thinks 'well that's a high dosage to me', 'well that's not to me'. Do you know what I mean? So, but that's difficult. The thing is, as a parent you hang on to every word. And I don't think that's what- I think that's maybe what they've forgotten. Em, so if a consultant comes round, I mean, he was days old and they put him on something, and one of the consultants turned around and said em, 'oh he's actually em, he shouldn't he shouldn't be on this now that he's born, this is a good drug for you know, pre-birth babies, but now that he's on this, this is actually poisoning him' and she says, and we went well 'what's the consequences?' and she said 'well, you know, em it can cause instant death'. And just kind of walked, you know! And we were like, but you hang on to <i>every</i> word, good or bad. I think you hang on to more bad stuff than good stuff. Cause if they say, he's doing great, you kind of go, 'phew', fine! But see if they say 'oh we don't know, it's not what we want', - you're suddenly:	Trying to make sense of why they get inconsistent treatment advice from various doctors in medical team. Trying to see it from medical team's perspective. Hang on to every word said by medical team- very sensitive to verbal communication; very important for medical team to be aware of this and be considered and sensitive in how they communicate with parents. Example of when a consultant communicated something verbally in an insensitive manner, leading to high levels of distress for parents without the consultant being aware of this. Parents still remember this incident even though it happened 6 years ago- significant impact of insensitive verbal communication on parents. Blunt use of words from consultant, leading to high levels of distress for parents. Consultant not aware of how this could affect parents, shown by way she just walked away after informing them the medicine can cause instant death. Parents remember the bad things that are communicated more than the good things- and the word-use stays with them. Importance of medical team being sensitive and considered in their word choice.
Impact of insensitive communication from the medical team		
Impact of insensitive communication from the medical team		
Impact of insensitive communication from the medical team		

<p>Living in a high state of anxiety</p> <p>Medical team forget what it's like for the parents</p> <p>Impact of insensitive communication from the medical team</p>	<p>'what do you mean it's not what you want? Is it really bad? ' you know, you do, you get like you're almost grabbing them and saying, and sometimes I think they maybe forget that, so they just rhyme off well 'd-d-d-d', you know. I mean I had a consultant one time, we'd been in for like over a week, and he came and had a word with me, and he said em 'and this one just doesn't seem to be working, I just, I just don't know what to do with him'. Do you know what I mean?! And I was just like, 'Oh my god! You don't know what to do with him! What are we gonna do with him?!' you know. And then of course, floods of tears and he was like, 'oh! Oh! Em, you know, no, but it's not that bad! We'll sort it, we'll sort it!' But that was his first thing, he was like that, you know, scratching his head: 'I don't know what to do with him'.</p> <p>I: He's just thinking out loud, and it's not helpful.</p> <p>G: the weird thing here is, that previous thing about the consultant mentioning that thing about the drug and it causing sudden death. Now this is where I can sort of sympathise with them, because in one respect I'm saying give us information. On the</p>	<p>Parents feel like consultants forget what it must feel like for them as parents- they are highly anxious about every word the consultant says, especially when the word implies there might be something wrong. Consultant sometimes might just be thinking out loud, but what they say deeply affects the parents.</p> <p>Example of when this happened.</p> <p>Parents have a very literal interpretation of what consultant says, when consultant might not intend for it to be understood that way.</p> <p>Example of when consultant realised the impact of his words on parents and backtracked, showing that he realised his word-use was not helpful. Seems that he did not realise the impact of his words until mum started crying- important for them to be more sensitive to cues from parents regarding their distress?</p> <p>Dad trying to see it from the consultant's position- showing the dilemma from their perspective; understand they're in a difficult position.</p>
<p>Seeing it from the consultant's perspective</p>		

Seeing it from the consultant's perspective	other hand, when the consultant does give information, we've been provoked (L laughing: we don't want that type of information!) So, I can understand why if, because the circumstances were not good, I mean he was like a day and a half old or something, we were shattered and you know, someone makes this off the cuff comment, and you'd (to L) been on this drug for months, and then someone says 'meh', and they're just giving you information. And so I can sympathise it is difficult for them, cause they're they're dealing with us who are obviously very concerned parents, and behind every word does become- and equally, the other issue we have is that, the thing that's been hardest, I suppose for any parents to get their head rounds is that there's a there's a huge area with [son] that is <i>unknown</i> . There's a whole unkno- they don't <i>know</i> . They don't <i>know</i> quite why this is happening. They don't actually quite <i>know</i> why these drugs are working.	Trying to explain it from parents' perspective again- circumstances "not good"- emphasising their difficult situation at the time and how this might have lowered their resilience to comments? Circumstances increase the distress caused by offhand comments due to lowered resilience.
Circumstances lower resilience to insensitive communication		
Seeing it from the consultant's perspective		Dad's use of "meh" to illustrate what consultant said- dad felt consultant did not put thought into what he said at the time? Again seeing it from consultant's perspective "just giving you information".
Living with uncertainty is the hardest part		Emphasising what mum said earlier- every word has meaning for them due to their high level of anxiety and concern.
		Hardest thing for them to deal with is the uncertainty surrounding son's condition- consultants don't have any answers, they don't understand son's condition well enough. I can sense the anxiety coming through his words.
Living with uncertainty is the hardest part	L: And they don't know <i>what's</i> going to happen, that's the point, they don't know whether the [gene mutation] is going to give him further problems or...or is this, is it going to stabilise at	Future is uncertain because of lack of knowledge regarding gene mutation. Living with uncertainty. Lots of unanswered questions about the future.

<p>Living with uncertainty is the hardest part</p>	<p>this?</p> <p>G: They don't know, it's genetic,</p> <p>L: And I think as a parent, you just want somebody just to kind of open a book and say 'this is what's gonna happen in the next 10 years and this is exactly how we're gonna play it, and it'll be fine'. And you're like, thank you. That's all you want to hear. But, you know, you don't get that, and it's hard.</p>	<p>Certainty and answers is what they want; even bad news about the future- if it is certain news- would be preferable to uncertainty. Really hard to not get any answers.</p>
<p>Living with uncertainty is the hardest part</p>	<p>G: If there was a distinction in how a consultant behaves in terms of when there's, if you break a leg and you've got a cast on they'll fix it in 6-8 weeks. I'm sure there'll be heart conditions that are like that. There will be, there are heart conditions, there are drugs, he'll go on this, stays on this, fine. It just means that this, this and this. With him [son] it's not like that. And you're living in this kind of perpetual em, sort of, em, it's a fog really. Em, not, fog's not the right word- you're em in a directionless map, do you know what I mean? You've got a map, there's nothing on it. We could go down this road, we could go down that road. We don't know how long that road will go for, it really is,</p>	<p>Dad comparing it to a physical health problem in which the answers and treatment is predictable. Wishes son's condition was like this. Doesn't mind if that would mean it had some negative implications, as long as they knew what to expect and what the treatment involved.</p> <p>Metaphor to explain what it feels like for them: change from describing it as a 'fog' to a directionless map. Maybe that they feel they can see clearly from their perspective but it's the medical team and the condition that isn't giving any answers, hence difference between fog and map?</p> <p>The future is uncertain; don't know what will happen and where they will end up. Constant sense of uncertainty and unknown.</p>

