

Trauma, growth and coping in parents of children with congenital heart disease:

A mixed methods study

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Abstract

Background: Raising a child with a congenital heart disease (CHD) can be a difficult experience. Posttraumatic stress symptoms have been observed in parents of children with CHD. However, there is little research investigating posttraumatic growth (PTG); or the positive changes following trauma, in this population. This study aimed to explore the experience of parents of a child with CHD as related to the constructs of traumatic stress symptoms and posttraumatic growth.

Method: A priority sequence approach was used with a preliminary quantitative survey. This was followed by asynchronous online discussion groups. Parents completed the: Psychological well-being posttraumatic changes questionnaire, Posttraumatic stress disorder checklist 5 and Perceived ability to cope with trauma questionnaire. Thematic analysis was used to analyse discussion group material.

Results: Parents reported experiencing symptoms of PTSD; and more than one fifth of parents scored above clinical cut offs for PTSD. The majority of parents reported positive change. Thematic analysis constructed three themes: being a parent of a child with CHD, facing uncertainty and the changes in our lives, which corroborate and extend previous findings.

Discussion: The implications of this study's findings are discussed in the context of previous research and the methodological considerations of the study. Clinical and research implications are also presented.

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Table 0.1 List of abbreviations

Abbreviation	Full construct
ADD	Adversity Activated Development model
APA	American Psychiatric Association
ASD	Atrial Septal Defect
BSI	Brief Symptom Inventory
BPS	British Psychological Society
CHD	Congenital Heart Disease
CoA	Coarctation of the Aorta
DCP	Division of Clinical Psychology
DSM	Diagnostic and Statistical Manual of the American Psychiatric Association
HLHS	Hypoplastic left heart syndrome
ICU	Intensive Care Unit
IQR	Inter-quartile Range
NHS	National Health Service
OHS	Open Heart Surgery
ONS	Office for National Statistics
PACT	Perceived Ability to Cope with Trauma scale
PCL-5	Posttraumatic stress disorder checklist -5
PICU	Paediatric Intensive Care Unit
PDA	Patent Ductus Arteriosus
PDS	Posttraumatic stress Diagnostic Scale
PMTS	Paediatric Medical Traumatic Stress
PTG	Posttraumatic Growth
PTSD	Posttraumatic Stress Disorder
PWB-PTC	Psychological wellbeing- Posttraumatic Changes questionnaire
SD	Standard deviation
SPSS	Statistical Package for the Social Sciences
TGA	Transposition of the Great Arteries
UK	United Kingdom
VSD	Ventricular Septal Defect

1. Introduction

1.1 Chapter Summary

This chapter defines congenital heart disease or defect (CHD), its prevalence, treatment and mortality and reviews the quantitative and qualitative research on the experiences of raising a child with CHD and the psychological distress experienced by parents. This is followed by a systematic literature review of the research on posttraumatic stress symptoms in parents of children with CHD. The construct of posttraumatic growth (PTG) is presented and the research on this topic in parents of children with CHD is reviewed. Finally the literature on coping in parents of children with CHD is discussed and the construct of coping flexibility is presented. The chapter concludes with the aims and research questions for the current study.

1.2 Congenital Heart Disease

1.2.1 Definition

The international statistical classification of diseases and health related problems manual (ICD-10, 1992) defines congenital heart disease (CHD) as malformations with the structure of the heart which are present at birth (World Health Organisation, 1992). Faults can involve the walls, valves or the blood vessels of the heart. CHD encompasses a diverse range of conditions which vary in severity and complexity. Conditions can be mild such as a small single septal defect; hole in the walls which divide the heart chambers. Moderately complex conditions can also occur where a number of different malformations are present at the same time, for example Tetralogy of Fallot. The most severe conditions occur where whole parts of the heart are missing or malformed, for example Hypoplastic Left Heart Syndrome (HLHS). Thus the diversity of conditions

under the term CHD, differ in complexity and severity and can give rise to a varied experience of the condition.

1.2.2 Prevalence

Heart defects are the most commonly occurring major congenital defect, (Dolk, Loane, & Garne, 2011; Springett et al., 2014; van der Linde et al., 2011) making the condition of particular interest. Estimates of prevalence rates of CHD can be complicated by a number of factors. Whilst the majority of CHDs are diagnosed in infancy (Wren & O'Sullivan, 2001), diagnosis can sometimes be outside of the data collection periods of epidemiological studies or national register reports and this can lead to underestimates of prevalence (Petersen, Peto, & Rayner, 2003). Cases can also be defined in different ways, that is some studies define cases in live births such as Wren and O'Sullivan (2001), whilst other register based studies and reports such as Springett et al. (2014) and Dolk et al. (2011) also account for cases amongst total births including: still births, late miscarriages and terminations of pregnancy due to foetal anomaly.

A meta-analysis of prevalence rates estimated 9.1 children were born with CHD per 1000 live births worldwide (van der Linde et al., 2011). In Europe the prevalence of CHD has been estimated between 7-8 per 1000 live births (van der Linde et al., 2011; Dolk et al., 2011). During the time period 1985–1999 based on the data from a single paediatric cardiac centre in a northern region of the UK, 5.2 cases of CHD were diagnosed in infancy per 1000 live births (Wren & O'Sullivan, 2001). After correcting for late diagnosis and under ascertainment Petersen et al. (2003) estimated the true prevalence of CHD in the UK to be 6.9 per 1000 live births. Another report estimated the birth prevalence of children with CHD in England and Wales for 2012 was 6.37 per 1000 total births (Springett et al., 2014). This report presented data from six regional areas and

estimated it accounted for 36% of total births in England and Wales. Of note neither of the reports by Wren and O'Sullivan (2001) or Springett et al. (2014) consider figures for areas such as London which have a higher than average maternal age and thus higher risk of congenital defects, potentially underestimating the true prevalence of CHD in England and Wales and the UK (Springett et al., 2014).

1.2.3 Treatment

Treatment of CHD is varied and depends on both the features of the defect, such as type and severity of the defect as well as the characteristics of the child such as: age, size, and current health. Not all forms of CHD require treatment; mild septal defects can heal on their own over time without intervention (Petersen et al., 2003). However, the more complex conditions can involve multiple surgical interventions, and in the most severe forms of CHD, such as HLHS, surgery is palliative and not corrective (Feinstein et al., 2012).

1.2.4 Mortality

The prognosis and mortality rate for children with CHDs varies considerably, dependent on the severity and complexity of the presenting heart condition (Petersen et al., 2003). Survival rates for the mildest conditions can be 100% (Petersen et al., 2003; Wren & O'Sullivan, 2001). For children with HLHS born during the period 1985–1994 in a single paediatric cardiology centre in the UK the 12 month observed survival rate was 0% (Wren & O'Sullivan, 2001). Due to improvement in surgical interventions for HLHS more recent survival rates have been estimated at 70% (Feinstein et al., 2012}.

Despite improvements in the treatment of CHD, of all infant deaths attributed to the impact of a congenital abnormality, heart defects remain the largest contributor, (Springett et al., 2014). The infant mortality rate related to CHD based on registers of congenital anomalies in England and Wales has been reported as 0.38 per 1000 live births in 2012 (Springett et al., 2014)

1.2.5 Current context of NHS CHD services

CHD services in the UK have been under scrutiny since the completion of the public inquiry into CHD care at the Bristol Royal Infirmary during the period 1984–1995 (NHS England, 2015). Despite this and subsequent reviews little coordinated change has taken place to improve CHD services. Most recently the NHS England board agreed the recommendations from the conclusions drawn in the NHS England’s two-year “New Congenital Heart Disease Review” (Huxter & Holden, 2015). This included: a new proposed model of care, service standards and specifications; changes to improve early diagnosis and information sharing; and proposals for commissioning, implementation, service monitoring and management. Currently NHS England is focusing on transitioning to commissioning and implementation of these new agreed standards to improve CHD services and patient experience. Within these new standards one key recommendation was for better support for patients and families including increased access to psychologists, among other health professionals. This has resulted in new standards specifying the requirement for the presence of practitioner psychologists experienced in working with CHD within each network of specialist children’s cardiology centres and specialist children’s surgical centres (NHS Board England, 2015). This shift acknowledges the growing literature reporting the psychosocial impacts of CHD on patients and their families. For psychologists working in this area, given the varied nature and treatment courses of CHD, in order to understand how to better support patient and

families' needs, it is important to understand the varied experiences of raising a child with the condition, and the potential psychological impact of treatment of the condition.

1.3 Experiences of parenting a child with a serious illness

The literature describing the experiences of parents of children with CHD forms part of the wider literature on the experiences of parents with children who have serious physical illnesses. Eiser (1993) describes two contrasting approaches within this literature: the prevailing medical position which stresses the differences between diseases and their idiosyncratic consequences, and a 'noncategorical approach' (Stein & Jessop, 1982) which emphasises the similarities across health conditions, in the demands chronic childhood illnesses places on children and their families. The former approach is manifest for example in disease specific journals and disease specific psychometric measures. This approach may also reflect the ways in which health professionals work in discrete and specialised teams with these conditions. One disadvantage of this approach has been described as the difficulty in then determining whether research findings are actually attributable to common factors across health conditions (Eiser, 1993). It might be equally argued that a 'noncategorical approach' might overlook important disease specific factors. Advocates of the 'noncategorical approach' have argued the expediency of this approach, given the low prevalence of some conditions and the similarities in the demands of chronic illness on parents. Eiser (1993) suggests that rather than the label attached to condition, what is important to understand is the degree of impact of the condition and its treatment on the family's ability to maintain everyday life.

Much of the qualitative research on the experiences of parents with a child with a serious illness is consequently disease specific. Some studies have selected parents on the

basis of characteristics of the child's current condition rather than diagnostic label, for example those needing complex respite care (MacDonald & Callery, 2008) or those transitioning to adult services (Heath, Farre & Shaw, 2017). A number of reviews across health conditions have been published including some meta-syntheses. Five review studies of parents' experiences across conditions including: three meta-syntheses (Bally, et al, 2018; Coffey 2006; Kepreotes Keatinge, & Stone, 2010), one rapid structured review (Smith, Cheater & Bekker, 2013) and one critical literature review (Fisher, 2001) are described in Table 1.1. The reviews used different definitions of serious and chronic illness and included studies which explored the experiences of parents with children with a wide range of different health conditions, for example: congenital conditions, cancer, progressive degenerative disorders, common conditions such as asthma and diabetes, Attention Deficit Hyperactive Disorder, and children who were technology dependent. The age of the affected child of parents in studies included the reviews ranged from birth to 46 years old indicating reviewed studies explored both current and retrospective accounts. The findings from each of these studies will be described in turn.

Fisher (2001) constructed the needs of parents who have children with chronic illness in three areas. The need for normality described a process of parents struggling to regain normality and control over uncertainty presented by the child's illness. The need for information described parents' constructions of information about their child's condition and treatment. The need for partnership described the process of negotiating parental involvement in the medical care of their child.

Table 1.0.1 Themes in review studies describing the experiences of parents with a child with a serious illness

Author, Year	Study	No. studies reviewed, Dates	Themes
Bally, et al. 2018	A meta-synthesis: uncovering what is known about the experiences of families with children who have life-limiting and life-threatening illnesses	23 1997-2014	+The devastation of living with uncertainty -Acknowledging a disrupted world -Reconciling with uncertainty +The emergence of hope -Changing priorities -Salvaging family relationships +Moving forward -Finding normal -Attaching new meaning
Coffey 2006	Parenting a child with chronic illness: a meta-synthesis	11 1989-2000	-Living worried -Staying in the struggle -Carrying the burden -Survival as a family -Bridge to the outside world -Critical times, -Taking charge.
Fisher, 2001	The needs of parents with chronically sick children: a literature review	8 1987-1997	-The need for normality and certainty -The need for information -The need for partnership
Kepreotes et al., 2010	The experience of parenting children with chronic health conditions: a new reality (Meta-synthesis)	10 2000-2006	-Sadness and chronic grief -Getting a grip -The need for information & support -Relationships with health professionals -Fragile control -A new reality
Smith et al, 2013	Parents' experience of living with a child with a long-term condition: a rapid structured review of the literature	34 1999-2009	-Parental impact -Illness management -Social context.

The study by Smith, Cheater & Bekker (2013) described three broad themes. “Parental impact” described parental responses such as the emotions experienced, how parents described the processes of adaptation, coping and the care giving role. The theme “illness management” described factors involved in parents providing medical and nursing care for their child. This included parents’ constructions of information and the process of acquiring knowledge and expertise in their child’s care and relationships with health professionals. “Social context” described the ways in which family life was changed by the child’s illness and parents’ positive and negative constructions of this. This study excluded studies with parents of children with: learning disabilities and those with terminal illness.

Coffey (2006) described seven themes across the reviewed studies common to the experience of parenting a child with a chronic condition. “Living worried” described the amount and breadth of parents’ current worries as well as their worries for the future of their child and the uncertainty associated with these worries. “Staying in the struggle” constructed “strife” in the experiences of parents. This included the experience of strong and overwhelming emotions such as anger, helplessness, hope and defeat, as well as coping strategies parents described using in managing their child’s care. “Carrying the burden” acknowledged the responsibility and impact on mothers as primary caregivers. “Survival as a family” described impact of the child’s condition on the whole family. This theme described impact on siblings, family routines and as well as parents’ worries about siblings’ wellbeing. “Bridge to the outside world” described the loss of freedoms, support systems and isolations parents experienced. This theme also described the importance of peer support groups, home care nurses and parents’ own spirituality in helping parents cope. “Critical times” captured specific significant events, their impact on parents and the

challenges to a fragile equilibrium of family life. This included: diagnosis, the first year after diagnosis, changes in condition and development milestones. Finally “taking charge” described parents developing an advocate role in their child’s care. This theme included relationships with professionals and strategies such as information seeking.

Six themes were constructed to a greater and lesser degree across the 10 studies reviewed by Kepreotes et al. (2010). The review by Coffey (2006) was amongst the two review studies included in this study, and thus some cross-over in themes is likely. “Sadness and chronic grief” was constructed as the common and overwhelming emotional experience of parents. “Getting a grip” acknowledged and described parents’ needs and attempts to gain control over the situations they faced. This included experiences of uncertainty and worry, and wide ranging strategies such as maintaining vigilance, coming to acceptance and deriving meaning from their experiences. “The need for information and support” described parents’ experiences of the medical model and their construction of the process of receiving and using information in caring for their child. “Relationships with health professionals” was related to the theme “the need for information and support” and described parents’ constructions of their role in their child’s medical care as well as positive and negative constructions of care received from health professionals. This theme touched on issues of power imbalance and constructions of the relationship with the healthcare professional. “Fragile control” described parents experiences of an outcome of the theme “getting a grip” and the needs for stability against a backdrop of inherent instability. Finally the theme “a new reality” acknowledged constructions of a changed reality across studies and described processes such as reframing and restructuring constructions of ways of living and ways of being a parent.

Bally et al., (2018) described the context of parents experiences as a “dual reality of death and survival” which was constructed as an oscillation between highs associated with survival and the lows associated with fear of their child’s death. Three themes and six subthemes were constructed to describe the processes in parenting a child with a life-limiting and life-threatening illness. The first major theme “the devastation of living with uncertainty” described the pervasive and overwhelming experience of uncertainty for parents and contained two subthemes. Firstly “acknowledging a disrupted world” constructed chaos and turmoil as a product of uncertainty, and described the intense emotions parents experienced in this process. The second subtheme “reconciling with uncertainty” constructed a process through which parents came to terms with their child’s condition and adapted to living with uncertainty. The second major theme “the emergence of hope” constructed the nature and value of hope as an essential resource in helping parents to endure the difficult times in their experiences. This theme was made up of “changing priorities”, which described as a reevaluation of priorities and values in life, and “salvaging family relationships”, which described the impact of the child’s condition on family relationships and emphasised positive changes in relationships resulting from parents’ experiences of their child’s health condition. The third major theme “moving forward” constructed a process through which parents survived and were transformed by their experiences and consisted of two sub-themes. Firstly “finding normal” was constructed as a process where parents sought to gain and maintain control over the situations they faced and to also see their child as normal. Finally “attaching new meaning” described parents constructing positive outcomes in a changed approach to life from their experiences.

Of the three meta-syntheses the two more recent studies, Bally et al, (2018) and Kepreotes et al (2010) described using Sandelowski and Barroso' (2006) methodology for data synthesis. Coffey (2006) described using Noblit & Hare's method (cited in Coffey 2006). Smith, Cheater & Bekker (2013) also clearly described using a systematic method of data synthesis based on the principles of thematic analysis. Whilst the study by Fisher (2001) reported issues of trustworthiness and rigour in the reviewed studies it did not specify the method of literature search or analysis in the critical literature review itself making it difficult to evaluate the soundness of the review.

Despite the differences in the five review studies they appeared to describe commonalities in the experiences of parents across health conditions. Each review constructed an overwhelming emotional experience in raising a child with a serious health condition and identified a major contributing role of uncertainty and worry in the experience. Most reviews described: a wider family systems impact, a key role of relationships with professionals, and parents' desires for information. Parents' constructions of positive changes were also noted in the majority of the reviews. It is likely that these themes can also be constructed in the experiences of parents of children with CHD and three studies which included the experiences of parents with children with CHD were incorporated in the reviews by: Bally et al., (2018); Coffey (2006), and Fisher (2001). However these studies were in a minority despite a broad and growing field of research in this area. This literature will be discussed next.

1.4. Experiences of raising a child with CHD

Parents often view the heart with more significance than other vital organs (Lok & Menahem, 2004; Wray & Sensky, 2004). The experience of raising a child with CHD is

also a broad research topic. Previous research, predominantly from a nursing perspective, has looked at specific experiences in raising a child with CHD such as receiving a diagnosis, surgical procedures and parenting. These will be discussed below.

1.4.1 Diagnosis

The impact of diagnosis of CHD on parents has been studied using quantitative methods. A cross-sectional survey of mothers who had received an antenatal diagnosis of CHD in their unborn child found pregnant women with foetuses with CHD had significantly higher anxiety (STAI) and depression (BDI-II) scores when compared with published norms of women with normal pregnancies (Rychik et al., 2013). These expectant mothers also reported high rates of traumatic stress symptoms; 39% of pregnant women scored above the clinical cut-off (Rychik et al., 2013). Similarly in a pilot study (Brosig, Whitstone, Frommelt, Frisbee, & Leuthner, 2007) also found that mothers diagnosed during the antenatal period reported significantly higher levels of distress, measured using the Brief Symptom Inventory (BSI), compared with norms. This difference was found at the time of diagnosis, birth, and 6 months after birth. Mothers of children diagnosed postnatally also reported significantly higher scores on the BSI of distress at the time of diagnosis which was found to have reduced 6 months after birth. Comparing the antenatal and postnatal diagnosis groups to each other, at diagnosis there was no difference in BSI scores however 6 months after birth the prenatal group had significantly higher BSI scores. However, severity of heart condition may have confounded this finding as the antenatally diagnosed group tended to have more severe forms of CHD than the postnatally diagnosed group (Brosig et al., 2007). Additionally, the sample sizes, $n=11$ and $n=16$ for the antenatal and postnatal diagnosis groups respectively were small. It was concluded that regardless of the timing, diagnosis was a distressing event for parents.

Qualitative methods have also been used to explore the retrospective experiences of parents receiving a diagnosis of their child's CHD. In a secondary analysis of interviews with eight parents using grounded theory analysis the experiences of parents of children with CHD were constructed as a transitional process through the following stages: something wrong, the illusiveness of normality, the rude awakening, managing uncertainty, new meanings, and taking stock (Messias, Gilliss, Sparacino, Tong, & Foote, 1995). The last two themes contained similarities with the concept of posttraumatic growth which will be discussed in section 1.6. In qualitative interviews with mothers of children antenatally and postnally diagnosed, Brosig et al. (2007) described the following common themes at diagnosis: feelings of; anger, disbelief, shock, guilt, and fear, difficulties parenting in the first 6 months, and the impact on the family. Those diagnosed postnatally also described wishing they knew sooner and those diagnosed antenatally described having time to prepare themselves. Receiving a diagnosis has been found to be a difficult time for parents.

1.4.2 Surgical procedures

Parents of children with CHD admitted for cardiac surgery have been found to have elevated levels of: stress, depression (Bevilacqua et al., 2013), parental stress related to hospitalisation (Franck, Mcquillan, Wray, Grocott, & Goldman, 2010) and reduced health related quality of life (Landolt, Buechel, & Latal, 2011). Whilst Bevilacqua et al (2013) found this did not differ according to antenatal or postnatal diagnosis, their group sizes were small (18 and 20) limiting the power of the study to find differences should they be present. As a pilot study the limited power of the study was acknowledged however no sample size calculation was provided. The proportion of parents of children with CHD reporting elevated levels of stress in one study were found to have decreased

from 63% to 25% one year after surgery (Wray & Sensky, 2004). This was similar to levels of stress found in the healthy comparison group, suggesting that stress levels in parents of children with CHD may reduce following surgery.

A number of qualitative studies have also examined parents' experiences of their child's cardiac surgery. Harvey, Kovalesky, Woods, & Loan (2013) used phenomenological analysis of journal entries from eight mothers to construct a model of "mothering through it all" with five main aspects: feeling intense fluctuating emotion, navigating the medical world, dealing with the unknown, facing the possibility of my baby dying, and finding meaning and spiritual connection. Also adopting a phenomenological approach Wei et al. (2016) analysed interviews with 10 mothers and 3 fathers post surgery. They described 10 key chronological experiences starting from receiving a diagnosis through to recovering from surgical procedures. In contrast Kosta et al. (2015) took a more pragmatic approach towards understanding parents' experiences with a view to improving parent's experiences. They used thematic and frequency analysis to examine structured interviews with 91 mothers and 63 fathers one month after hospital discharge. They used a top down framework of helpful aspects, difficult aspects and aspects parent would have liked to be different, to organise their data. Despite the varied time points of data collection, from one week post surgery (Wei, et al., 2016) to up to 4 years post surgery (Harvey et al., 2013), there were a number of similarities between the themes developed in the studies by Wei et al.(2016) and Harvey et al. (2013) which included: difficult and changing emotions, uncertainty or the unknown, finding meaning and benefit finding in the experience, self blame for the child's condition, and difficulties with medical decision making. Similarly the study by Kosta et al. (2015) which also used a current inpatient sample also highlighted the "uncertain and unfolding nature of the

diagnosis and surgery” (p1059). The studies by Kosta et al. (2015) and Harvey et al. (2013) both identified support and the wider systems as themes. Clearly there are similarities in the themes parents described as part of their experiences at diagnosis. There were also differences in some of the themes constructed. One theme came from the view of fathers and described a dual concern for the child and for their partner (Wei et al., 2016). Only the study by Harvey et al. (2013) explicitly described facing the possibility of the child dying. Whilst parents’ experiences of their child’s cardiac surgery were constructed as challenging; emotionally, psychologically and practically, it is interesting that the two phenomenological studies described themes in which parents expressed finding meaning or benefit in their experiences. This could be again considered a form of posttraumatic growth which will be discussed in section 1.6.

The recent review of CHD services has highlighted an overemphasis on surgery as a means to improving CHD services (NHS England Board, 2015). This suggests that broader issues, such as parenting a child with CHD, should also be considered in understanding parents’ experiences.

1.4.3 Parenting

Several qualitative studies have explored parenting a child with CHD. Three studies conducted by Rempel’s research group in Canada have used grounded theory analysis to examine the experience of parenting children with HLHS, a severe form of CHD with only palliative surgical treatment (Rempel & Harrison, 2007; Rempel, Rogers, Ravindran, & Magill-Evans, 2012; Rempel, Ravindran, Rogers, & Magill-Evans, 2013). In their first study they described “extraordinary parenting” in a context of uncertainty and a three-part process of “safeguarding precarious survival” (Rempel & Harrison,

2007). This involved safeguarding survival of the child, the self and the couple. Following advances in the surgical treatment of children with HLHS (Rempel et al., 2013) used a similar methodology, expanding the informant groups to include grandparents based on their finding in their earlier study, to again examine the experiences of parenting a child with HLHS. Parents and grandparents were asked to recall their experience from the time of the child's diagnosis to the present. From this data they constructed a four stage process model of "parenting under pressure" which consisted of: realising and adjusting to the inconceivable, growing increasingly attached, watching for and accommodating to the unexpected and encountering new challenges (Rempel et al., 2013). Based on a secondary analysis of the same data they constructed a complementary five aspect model of parenting a child with HLHS consisting of: survival parenting, hands off parenting, uncertain parenting, expert parenting and supported parenting. Again in the study by Rempel et al. (2012) parents constructed a narrative of personal growth from their experiences which could be considered an aspect of posttraumatic growth.

A study by (Moola, 2012) used a narrative approach to thematic analysis to compare the experiences of parents of children with CHD to parents of children with cystic fibrosis. They described three themes, two of which related to the experiences of parents of children with CHD: "the stress of parenting a child with a chronic illness" and "learning to put things into perspective". Using Frank's (1995) illness typology the study concluded that the experiences of parents of children with CHD could be characterised by a drawn out restitution narrative of a period of ill health, convalescence and an eventual return to health. In contrast the experiences of parents of children with cystic fibrosis were characterised by chaos and quest narratives of unresolved feelings of loss of control

and engulfment by illness; and transformation of the body and the self and reprioritisation of values. However this finding may be to some degree related to certain features of the sample. Firstly the severity of the children's heart condition was not stated although it is reported that some children had never been hospitalised it is unclear which parent group this refers to. Additionally the age of the children at the time of data collection may be significant as the children were aged between 10 and 18 and it is likely that cystic fibrosis and CHD have different illness courses which impact on parents differently at different points in time. As most surgical procedures for CHD occur within the first 5 years of life the parents of children with CHD in this sample would have had more time passed since any surgical procedures and this may have affected their responses.

In summary previous research has demonstrated the experiences of: diagnosis, surgical procedures and parenting a child with CHD are challenging and associated with adverse impact on parents. A number of studies have specifically investigated the incidence of psychological distress and traumatic stress symptoms in parents of children with CHD in their responses to these challenging and distressing events.

1.4.4 Psychological distress in parents of children with CHD

Many studies have examined the mental health of parents of children with CHD more generally. A review of 25 studies highlighted the degree of stress faced by parents raising a child with CHD and concluded parents demonstrated elevated rates of stress, worry, anxiety and depression (Soulvie, Desai, White, & Sullivan, 2012). Another recent systematic review of research on families of children with CHD also concluded that 75% of reviewed articles investigating parents' psychological health reported higher levels of stress, anxiety and depression compared with the general population (Wei, Roscigno,

Hanson & Swanson, 2015). The remaining 25% of reviewed studies reported no difference or lower levels of psychological distress compared with the general population. It was suggested that the discrepancies may be due to the use of different measures of distress (Wei et al., 2015). Despite the discrepancies in findings, psychological distress in parents of children with CHD is an important outcome to monitor and reduce as it has been found to impact negatively on child behaviour and functioning (Landolt, Ystrom, Stene-Larsen, Holmstrom, & Vollrath, 2014; Spijkerboer et al., 2007). Similarly posttraumatic stress symptoms in parents have been found to be related to poorer recovery from posttraumatic stress symptoms in hospitalised children (Landolt, Ystrom, Sennhauser, Gnehm, & Vollrath, 2012).

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1.5 Posttraumatic stress disorder (PTSD) and symptoms

According to the current Diagnostic and Statistical Manual of the American Psychiatric Association (APA) (DSM-5, 2013) PTSD is now classified as a trauma and stressor related disorder closely related to anxiety disorders rather than a subtype of anxiety disorder (APA, 2013). A diagnosis of PTSD is based on eight diagnostic criteria. The primary diagnostic criterion for PTSD, or criterion A, is exposure to a traumatic event, with a response characterised by symptoms in each of the four symptom criterion clusters of: intrusion, avoidance, negative changes in cognition and mood, and changes in arousal and reactivity. The last three criteria specify the symptoms have duration of longer than a month, cause clinically significant distress or impairment in functioning, and finally these symptoms are not caused by substance use or another condition. Population prevalence rates based on the new DSM-5 criteria have not yet been published. The projected lifetime prevalence of PTSD, based on DSM-IV criteria, has been estimated at 8.7% in the United States (APA, 2013). The risk of developing PTSD

based on DSM-III-R criteria following a traumatic event was found to be 8.1% for men and 20.4% for women (Kessler, Sonnega, Bromet, Hughes, & Nelson, 1995). Higher prevalence rates have consistently been found in women and this is beyond differences in the type of traumatic event experienced by men and women (Kessler et al., 1995). Female gender is thought of as a risk factor for PTSD (APA, 2013), suggesting that mothers are more at risk than fathers.

In the UK the 2014 Adult Psychiatric Morbidity Survey in England reported 4.4% of respondents met DSM IV criteria for PTSD in the last month using the PTSD Checklist (Civilian version) (McManus, Jenkins & Brugha, 2016). This is much lower than US estimates. It was suggested that this estimate may underestimate the prevalence of PTSD in some communities due to differences in exposure to traumatic events which differs according to geographical and demographic factors (Frissa, Hatch, Gizard, Fear, & Hotopf, 2013). In an inner city UK community sample the prevalence of current PTSD symptoms was estimated at 5.5% using the Primary care PTSD screen (Frissa et al., 2013).

1.5.1 Criterion A

The definition of the critical PTSD criterion A stressor which defines what may be considered a traumatic event has changed over the years with each revision of the DSM, from the first appearance of the disorder in the DSM-III in 1980 to the current DSM-5. The current PTSD Criterion A states that the person must have been exposed to: “actual or threatened death, serious injury or sexual violence” in one of four ways: “directly, witnessing in person; learning the event occurred to a close family member or friend; or experiencing repeated or extreme exposure to aversive details of the traumatic event(s)”

p271 (APA, 2013). Under the new criteria, if a traumatic event happened to a close family member or friend and the event involved actual or threatened death, “the event(s) must have been violent or accidental” p 271 (APA, 2013). To qualify, medical incidents must involve “sudden, catastrophic events” p274. The exception to this is for indirectly experienced events, witnessing “a medical catastrophe in one’s child” p274 is included as a traumatic event. However what is experienced as a catastrophe is subjective and is likely to vary between medical experts and parents and may also vary between parents. The threat to the life of the child during and following major cardiac surgery, witnessing painful invasive medical procedures carried out on their child and exposure to the experiences of other children in the hospital setting, might be considered traumatic events by parents. Whereas for medical professionals with experience in this area, risk and threat to life may be evaluated differently.

1.5.2 Posttraumatic stress symptoms in parents of children with physical health problems.

A range of posttraumatic stress symptoms such as: physical arousal, re-experiencing and avoidance have been measured in parents of children with physical health problems. For example: parents of infants admitted to neonatal and paediatric intensive care (Balluffi et al., 2004; Lefkowitz, Baxt, & Evans, 2010; Shaw et al., 2006), parents of children admitted to hospital (Wray, Lee, Dearmun, & Franck, 2011) and parents of childhood cancer survivors compared to healthy controls (Kazak et al., 1997) Parents of children with congenital anomalies have also described their experiences of receiving a diagnosis as traumatic (Aite et al., 2011) and have been found to have high levels of PTSD (Horsch, Brooks, & Fletcher, 2013). Traumatic stress symptoms have also been observed in parents of children with CHD and will be described in the next section.

1.5.3 Posttraumatic stress symptoms in parents of children with CHD

A systematic search of the literature was conducted with the aim of ascertaining the current findings on the prevalence and experience of posttraumatic stress symptoms and PTSD in parents of children with CHD.

1.5.3.1 Literature Review

1.5.3.1.1 Search strategy.

The following electronic databases were searched: PsycINFO, Psychology and Behavioral Sciences Collection, PsycARTICLES, MEDLINE, CINAHL with Full Text, for articles published from the earliest date for each database, until April 2016. The following search terms were used in combination with Boolean operators: ‘congenital’ AND (‘heart defect’ OR ‘heart disease’ OR ‘cardiac’ OR ‘anomaly’ OR, ‘lesion’ OR ‘malformation’) AND (‘parent*’ OR ‘mother*’ OR ‘maternal’ OR ‘father*’ OR ‘paternal’ OR ‘caregiver’) AND (‘posttraumatic stress’ OR ‘posttraumatic stress’ OR ‘posttraumatic stress’ OR ‘traumatic stress’ OR ‘acute stress’ OR ‘stress reaction’ OR ‘PTSD’). The search was limited to articles published in English. The Cochrane library database was also searched and the abstracts from the reference lists of the included studies were screened for additional relevant publications.

1.5.3.1.2 Article selection.

All identified abstracts were initially screened to assess whether the full article would be obtained. Theoretical papers, and unpublished theses and dissertations were excluded. Full articles were then evaluated to determine whether the study met inclusion

criteria. The primary inclusion criterion was that the study participants included parents of children with CHD. A second inclusion criterion required that the study investigated traumatic stress symptoms. A third inclusion criterion was that children of study participants were under the age of 18.

1.5.3.1.3 Search results

The electronic database literature search identified 17 abstracts. One further article was found from reference list search. From these, eight abstracts met inclusion criteria and full text articles were obtained. Following this two further studies were excluded: one study reported finding from parents of children with congenital anomalies and did not separate out findings for parents of children with CHD (Horsch et al., 2013) and another study because the study did not separate the findings from parents of children with CHD from the findings from parents of children with cancer (Burke et al., 2014). Thus six studies were reviewed (see Appendix A for flow diagram).

1.5.3.1.4 Data extraction

The data extracted from quantitative articles included: study design, sample and sample size, child age at the time of data collection, time of data collection, traumatic stress measure used and the main study findings.

1.5.3.1.5 Article summaries and characteristics

Five quantitative studies and one qualitative study were found which reported traumatic stress symptoms in parents of children with CHD. A summary table of the study characteristics is presented in Table 1.1 below which shows that the studies varied in

nearly all aspects assessed. Helfricht, Latal, Fischer, Tomaske, & Landolt, (2008) conducted a prospective cohort study to examine PTSD in parents of children with CHD undergoing cardio-pulmonary bypass surgery. They assessed PTSD at discharge following surgery and six months after discharge. Helfricht et al. (2009) used a cross-sectional survey to conduct a psychometric evaluation of a German version of the Acute Stress Disorder Scale in parents of children with CHD following cardiopulmonary bypass surgery and an adult post-surgical cardiac sample. The studies by Franich-Ray et al. (2013) Toren & Horesh, (2007) & Rychik et al. (2013) used cross-sectional survey design to examine traumatic stress symptoms in parents of children with CHD, at different time points. The study by Franich-Ray et al. (2013) aimed to investigate the prevalence of trauma symptoms in parents of infants who had cardiac surgery before the age of 3 months. They assessed acute stress one month after hospital discharge. The study by Rychik et al. (2013) aimed to determine whether prenatal diagnosis of CHD increased maternal stress. They assessed traumatic stress symptoms, depression, anxiety and coping in mothers two to four weeks after prenatal diagnosis. The study by Toren & Horesh (2007) aimed to assess psychiatric morbidity in adolescents who had childhood operations for cyanotic heart disease and their parents. They screened for traumatic stress symptoms, anxiety and depression in adolescents and their parents at follow up, 13.7 ± 2.48 years ($\bar{x} \pm SD$) after cardiac surgery. The qualitative study by Re, Dean, & Menahem (2013) used semi-structured interviews to explore the experiences of mothers of children with CHD who required surgery in infancy. Systematic content analysis was used to understand the data. The quantitative studies will be evaluated together first.

Table 1.2 Summary of study characteristics for studies identified in a literature search of PTSD in parents of children with CHD

Authors	Country, Design	Sample	Participants	Age of child at time of data collection	Time of data collection	Traumatic stress measures	Main Findings
Franich-Ray et al., 2013	Australia, Cross-sectional survey	Parents of children having cardiac surgery before the age of 3 months	77 Mothers 57 Fathers	\bar{x} = 51.7 days SD= 42.07	1 month after hospital discharge	Acute Stress Disorder scale	27% of parents scored above the cut-off for ASD No significant differences between parents Dissociative symptoms most commonly endorsed
Helfricht et al., 2008	Switzerland, Prospective cohort study	Parents of children having cardiopulmonary bypass surgery	122 Mothers 92 Fathers	0-16 yrs	At discharge from hospital and 6 months after discharge	Posttraumatic Diagnostic Scale (German)	At discharge 16.4% of mothers and 13.3% of fathers scored above the clinical cut-off for acute PTSD 6 months after discharge these were 14.9% and 9.5% respectively. No significant differences between parents Acute symptoms predicted 6 month outcomes.
Helfricht et al., 2009	Switzerland, Cross-sectional survey – psychometric evaluation of measure	Parents of children having cardiopulmonary bypass surgery Post surgical cardiac patients	35 Mothers 26 Fathers 38 Men 14 Women	0-16 yrs N/A	\bar{x} = 22 days (SD= 7) after surgery 1 wk after transfer from ICU	Acute Stress Disorder scale (German)	25% of parents scored above the cut-off for ASD 4% of cardiac sample scored above the cut-off for ASD

Authors	Country, Design	Sample	Participants	Age of child at time of data collection	Time of data collection	Traumatic stress measures	Main Findings
Toren & Horesh 2007	Israel, Cross-sectional survey	Adolescents who had past cardiac surgery for CHD correction and their parents	22 Mothers 9 Fathers	\bar{x} = 14.7yr, SD= 0.3 Child age at time of surgery \bar{x} =1.8yrs, SD= 1.9	At cardiac follow-up on average 13.7 yrs after surgery (SD=2.48)	Posttraumatic Diagnostic Scale	3% of parents met criteria for full PTSD 12% of parents met criteria for partial PTSD
Rychik et al., 2013	USA, Cross-sectional survey	Expectant mothers receiving a diagnosis of CHD for their unborn child	59 Mothers	Unborn child	2-4weeks after diagnosis	Impact of Events Scale	39% of mothers scored above the clinical cut off for PTSD Significant relationships were found between depression, anxiety and PTSD and coping
Re et al., 2013	Australia Qualitative interviews	Parents of children admitted for cardiac surgery	26 Mothers	2 months old	After surgery	Thematic content analysis	Acute stress in the most difficult experience Diagnosis Traumatic separations Experiences of ICU

1.5.3.2 Evaluation of quantitative findings

Prevalence of traumatic stress symptoms

Prevalence rates of traumatic stress symptoms varied considerably between the studies. Helfricht et al. (2008) reported at discharge from hospital following surgery 16.4% of mothers and 13.3% of fathers met symptom criteria for PTSD, based on DSM-IV. This prevalence was found to decrease slightly but significantly 6 months after discharge (14.9% of mothers and 9.5% of fathers). PTSD severity at discharge was found to predict severity at 6 months. Using the same measure Toren & Horesh (2007) found that at follow up many years after surgery 3% of parents met criteria for PTSD. Both these studies reported lower prevalence rates than Franich-Ray et al. (2013) found in parents of children with CHD one month after hospital discharge. Franich-Ray et al. (2013) reported 27% of parents met criteria for Acute Stress Disorder, according to DSM-IVR criteria. Using the same measure Helfricht et al. (2009) found 25% of parents met criteria post-surgery. Rychik et al., (2013) used the Impact of Events Scale to determine that the prevalence rate of PTSD in mothers two to four weeks after prenatal diagnosis of CHD was 38.9%. It appears that PTSD symptoms decrease with increasing time however the different measures used and the different time of assessment in these studies make it difficult to conclude what the differences in these prevalence rates reflect. Additionally the different time points of assessment and different potentially traumatic events referenced in the study, point to the question of the potential cumulative effect of a number of potentially traumatic events on parents' experience of traumatic stress symptoms.

The study by Toren & Horesh (2007) reported PTSD prevalence rates lower than UK population estimates of PTSD (see section 1.4). This may have been because a

large number of years had elapsed since the child's surgery; it is possible that parents received treatment or their symptoms spontaneously remitted during this time. However a number of methodological issues may have also contributed to this finding. For example the smaller sample size may have underpowered the study; however no sample size calculation was reported. Additionally issues with selection bias in the sample may have contributed to underestimation of PTSD symptoms. The response rates and participant selection procedure are not described in the study making it difficult to assess potential sources of selection bias. Compared to the sample in both the Franich-Ray et al. (2013) and Rychik et al. (2013) studies participants in the Toren & Horesh, (2007) study had a much higher level of education. Education is often used as a proxy measure for socioeconomic status (SES). Both higher SES and higher educational attainment have been found to be protective factors against PTSD (APA, 2013).

As cross-sectional studies assess exposure and outcome at a single point in time, they provide descriptive information and cannot be used to determine causality (Churchill, 2003). Thus, whilst the studies by Franich-Ray et al., (2013) Helfricht et al., (2009) and Rychik et al.(2013) showed much higher prevalence of traumatic stress symptoms in parents of children with CHD compared to UK general population prevalence estimates (see section 1.4) it cannot be concluded that it was the child's CHD that caused a higher prevalence of PTSD.

Acute stress disorder and PTSD

The symptom profiles of Acute Stress Disorder and PTSD are almost identical with two exceptions. Firstly the duration of Acute Stress Disorder is from 3 days to 1 month after the traumatic event, whilst PTSD is diagnosed after one month. Secondly

diagnosis of Acute Stress Disorder places more emphasis on dissociative symptoms. Acute Stress Disorder was introduced into DSM-IV with the aim of distinguishing between trauma survivors who would recover spontaneously over time from those who would not and thus show those who would benefit from earlier intervention (Cahill & Pontoski, 2005). Although the pathologising of what might be a normal and temporary state was not without controversy (Harvey & Bryant, 2002).

The above studies assessed a mix of Acute Stress Disorder and PTSD. The initial assessment at time of discharge in the Helfricht et al.(2008) study could be considered to assess Acute Stress Disorder rather than PTSD as DSM-5 PTSD criterion F states the symptom duration should be greater than one month (APA, 2013). The symptoms parents reported may have been present for less than a month if surgery was considered as the traumatic event. Similarly if diagnosis was considered the traumatic event in the Rychik et al. (2013) study then they may have assessed Acute Stress Disorder rather than PTSD. Additionally the study by Helfricht et al. (2009), which used the Acute Stress Disorder scale to assess symptoms on average 22 days after surgery, also assessed Acute Stress Disorder rather than PTSD if surgery was taken as the traumatic event. Whilst measures of Acute Stress Disorder are still relevant, higher prevalence rates would be expected if assessing Acute Stress Disorder compared to PTSD as participants have not yet had time to process the trauma and may still be in the initial period of experiencing symptoms of intrusions and avoidance. Whilst it is highly likely that those meeting criteria for Acute Stress Disorder go on to meet criteria for PTSD it is not inevitable (Cahill & Pontoski, 2005).

Other potential sources of bias

The overall response rate in the Franich-Ray et al.(2013) study was low (38.8% and 27.7% of the total population of mothers and fathers respectively), indicating a potential source of bias in the sample. More than one fifth of potential participants were excluded because their child was considered too ill or had passed away. Although Franich-Ray et al.(2013) reported high prevalence rates of PTSD, this would suggest that parents with the most difficult experiences were excluded from the study and this may have underestimated levels of traumatic stress symptoms.

The response rates of 61% and 56.1%, reported in the two data collection points of the Helfricht et al. (2008) study whilst higher than those reported by Franich-Ray et al. (2013) indicate a potential source of selection bias in the study. Significant differences were found between the eligible patient families for the study who declined to participate and study participants. Helfricht et al. (2008) reported some reasons given for non-participation as “too strained” and “demanding/ difficult family situation”, this could indicate higher levels of distress and potentially traumatic stress symptoms. Parents who declined to participate in the study were found to be of significantly lower SES. SES has been described as a risk factor for PTSD (APA, 2013) thus potentially underestimating levels of PTSD in parents of children with CHD.

1.5.3.3 Symptoms of PTSD in qualitative themes

In the study by Re et al. (2013) 26 mothers of infants with CHD were recruited from the local population admitted to a cardiac surgical centre. At the time of the interview infants had undergone cardiac surgery and were two months old. Re et al. (2013) reported “almost all mothers described symptoms of acute stress and posttraumatic

stress symptoms” p 280 (Re et al., 2013). Mothers in the study described: feelings of dissociation, helplessness, horror, anxiety, and confusion. This description fits with DSM-IV PTSD criterion A2 requirement for responses to the traumatic event to involve feelings of “fear, helplessness and horror” p 467 (APA, 2000), however this requirement has been removed from DSM-5 (Friedman, 2013). This decision was made based on two rationales. Firstly some trauma survivors, such as military personnel, did not report this subjective experience when facing traumatic events and secondly this criterion was not predictive of PTSD. It was thought that this change in criteria would lead to increases in PTSD prevalence as those who previously did not meet criteria based on this criteria now would, according to the new criterion (Friedman, 2013).

Re et al. (2013) reported two main themes: “acute stress in the most difficult experience” and “reflections on mothers’ experiences of participating in the research”. The first theme highlighted traumatic events and parents’ emotional and psychological responses and consisted of three subthemes: diagnosis, traumatic separations, and experiences of ICU. Diagnosis was experienced as time of heightened stress by all mothers. Traumatic separations were also described as a common experience. These difficult separations included separation at birth and for surgery. Feelings of anxiety whilst waiting during surgery and not knowing the outcome were described. Finally experiences of the “horror” of ICU, medical procedures were described as generating acute stress for mothers. In the second theme one of the subthemes “containing the chaos of trauma” Re et al. (2013) described some mothers noted difficulty in talking about and reflecting on their experiences. Re et al. (2013) concluded that the opportunity that the research interview presented to process and integrate what parents’ had been through was experienced as therapeutic.

Evaluating qualitative rigour

Evaluation of the paper by Re et al. (2013) showed the researchers had considered the elements outlined in the Critical Appraisal Skills Programme for quantitative and qualitative research (CASP, Public Health Resource, 2013). The authors described the study context, methods of data collection and analysis, improving the dependability of the findings. Researcher reflexivity and transparency of researcher positioning were acknowledged and some discussion with co-authors and colleagues was described in developing themes, supporting confirmability of the findings. Direct quotes were used to evidence the themes, enhancing the credibility of the findings. For a number of potential participants information was missing, precluding inclusion in the study suggesting a potential threat to transferability of the findings. On the whole the paper was judged as sound.

1.5.3.4 Summary of literature review

The literature search found six studies addressing the prevalence and experience of traumatic stress symptoms in parents of children with CHD. Prevalence rates of PTSD ranged from 39% of mothers assessed 2-4 weeks after prenatal diagnosis (Rychik et al., 2013) to 3% of parents assessed on average 13.7 years after surgery (Toren & Horesh, 2007). Whilst suggestive of a decrease in PTSD symptoms over time, prevalence estimates assessed both PTSD and Acute Stress Disorder based on DSM-IV and DSM-IVR criteria at different time points in the child's illness trajectory and the studies also used a number of different measures making direct comparison difficult. No prevalence estimates were reported for parents of children with CHD in UK populations. PTSD prevalence estimates in the general UK population have varied from 4.4% (McManus et al., 2016) to 5.5% (Frissa et al., 2013).

The theme of “Acute stress in the most difficult experience” developed by Re et al. (2013) described symptoms of dissociation and helplessness which corresponded to symptoms of PTSD and support the quantitative findings of traumatic stress symptoms in parents of children with CHD. However in addition to the specific issues as to how DSM diagnostic criteria for PTSD might be applied to parents of children with CHD discussed above there are also wider critiques about psychiatric diagnoses more generally which are relevant in understanding the context behind, and the meaning of these findings.

1.5.3.2 Current critiques of functional psychiatric diagnoses

Psychiatric diagnosis has been criticised on many fronts in terms of its usefulness, validity, reliability, cultural bias, and the negative impact on those who receive a diagnosis (Bentall, 2004, Johnstone, 2014; Kirk & Kutchins, 1997). These arguments have tended to focus on “functional diagnoses” such as schizophrenia, bi-polar disorder, personality disorder etc. It has been argued that the persistence of the use of psychiatric diagnosis despite these longstanding criticisms is because this criticism strikes at the very core of the epistemology of the nature of mental distress, the application of a ‘disease’ or medical model in the construction of “mental illness” and the legitimisation of the role of the medical profession in its treatment (Johnstone, 2018). Coinciding with the release of the latest revision of the DSM, the British Psychological Society’s (BPS) Division of Clinical Psychology (DCP) launched a position statement on the classification of an individual’s behaviours and experiences in terms of functional psychiatric diagnoses (DCP, 2013), in which they acknowledged the limitations of functional psychiatric diagnoses despite their current standing in mental health services, the media, and the public understanding. The DCP (2013) called for a “paradigm shift” away from ‘disease’ model conceptualisations and towards multi-factorial approaches which acknowledged

complexity and centred the core principles of psychological formulation in contextualising distress and behaviours, in order to make sense of the individual's behaviours and experiences. Whilst these differences in conceptual and philosophical frameworks might appear due to a professional divide between psychiatry and psychology, many have emphasised it is not (critical psychiatry network, 2013, cited in Johnstone, 2018). In a similar vein a number of psychiatrists also called for psychiatry to move beyond a "technological" paradigm which they described as positivistic, based on a conceptualisation of mental health problems caused by abnormal or faulty physiological or psychological processes within the individual, processes which operate independently of context (Bracken et al., 2012). Examples of approaches from this paradigm might also extend to disorder specific Cognitive Behavioural Therapy (CBT) treatment models based on a premise of disordered or abnormal functioning of cognitive processes.

Hard stances in this ongoing debate have polarised psychiatric diagnosis against psychological formulation in an epistemological dilemma, suggesting that the two contradictory explanations cannot coexist (Johnstone, 2018). Others have described the benefits and challenges in interprofessional collaboration which tolerates these differences in positioning on psychiatric diagnosis, and opens up space for dialogue (Tan & McConvey, 2014). Whilst these critiques have focussed on functional diagnoses there are also implication for the diagnosis of PTSD and additional critiques of the construct of PTSD as a disorder.

1.5.3.3 Current critiques of the construct of PTSD

Critiques of PTSD have argued that rather than representing a "disease" PTSD is as much a construct of sociopolitical ideas from a particular time in history (Summerfield,

2001). Summerfield (2001) described the origins of the label PTSD in the experiences of American soldiers in the aftermath of the controversial Vietnam war and proposed that the new disorder allowed them a victim as opposed to a perpetrator role in the hostilities, and along with this because PTSD was constructed as a medical disorder, it conferred a sick role which gave certain advantages; for example rights to disability pension (Summerfield, 2001). Thus it can be argued that the construct of PTSD created space for the creation of new roles and new narratives around the experiences of these soldiers. This view is based on critique of the social construction of PTSD as a disease or disorder rather than questioning whether PTSD describes experiences of suffering or distress. Summerfield (2001) highlights the modern “conflation of distress with “trauma” (p96) and the pervasive use of trauma as a modern idiom of distress. Amongst other critiques of the construct of PTSD as a disorder are that: it pathologises a natural response to events, it is based on unreliable self-report accounts of the criterion A stressor and subsequent symptoms, and that it describes a culture bound syndrome (Friedman, Resick & Keane 2007).

The position of this study is that whether traumatic events meet the criteria for a psychiatric diagnosis or not and whether posttraumatic stress symptoms reach clinical thresholds or not, the distressing experiences these symptoms represent, and the context and meaning these experiences have for parents warrant consideration. The research literature suggests that parental traumatic stress symptoms have been found to predict child traumatic stress symptoms (Nugent, Ostrowski, Christopher, & Delahanty, 2007). There is also evidence to suggest that maternal traumatic stress symptoms increase the risk of the development of insecure mother-infant attachment styles and that insecure attachment styles influence the severity of any PTSD experienced later in life (Bosquet

Enlow, Egeland, Carlson, Blood, & Wright, 2014). Whilst more research is needed to elucidate whether attachment mediates the intergenerational transmission of PTSD, insecure attachment has been associated with other adult mental health problems for example depressive symptoms (Simpson & Rholes, 2004) and post-natal depression (Bifulco et al., 2004). Subclinical traumatic stress symptoms in parents might also interfere in treatment seeking behaviours for their children, information processing and medical decision making. Thus it is important to recognise and assess the difficulties parents experience so that appropriate support can be offered to parents and children. Understanding the distressing experiences is the first step in supporting parents who need help coping. Whilst the distressing experiences described by parents are often described in the literature as traumatic stress symptoms and PTSD these experiences and the context and development of these experiences can also be understood in terms of a specific contextual model of Paediatric Medical Traumatic Stress (Kazak et al., 2006).

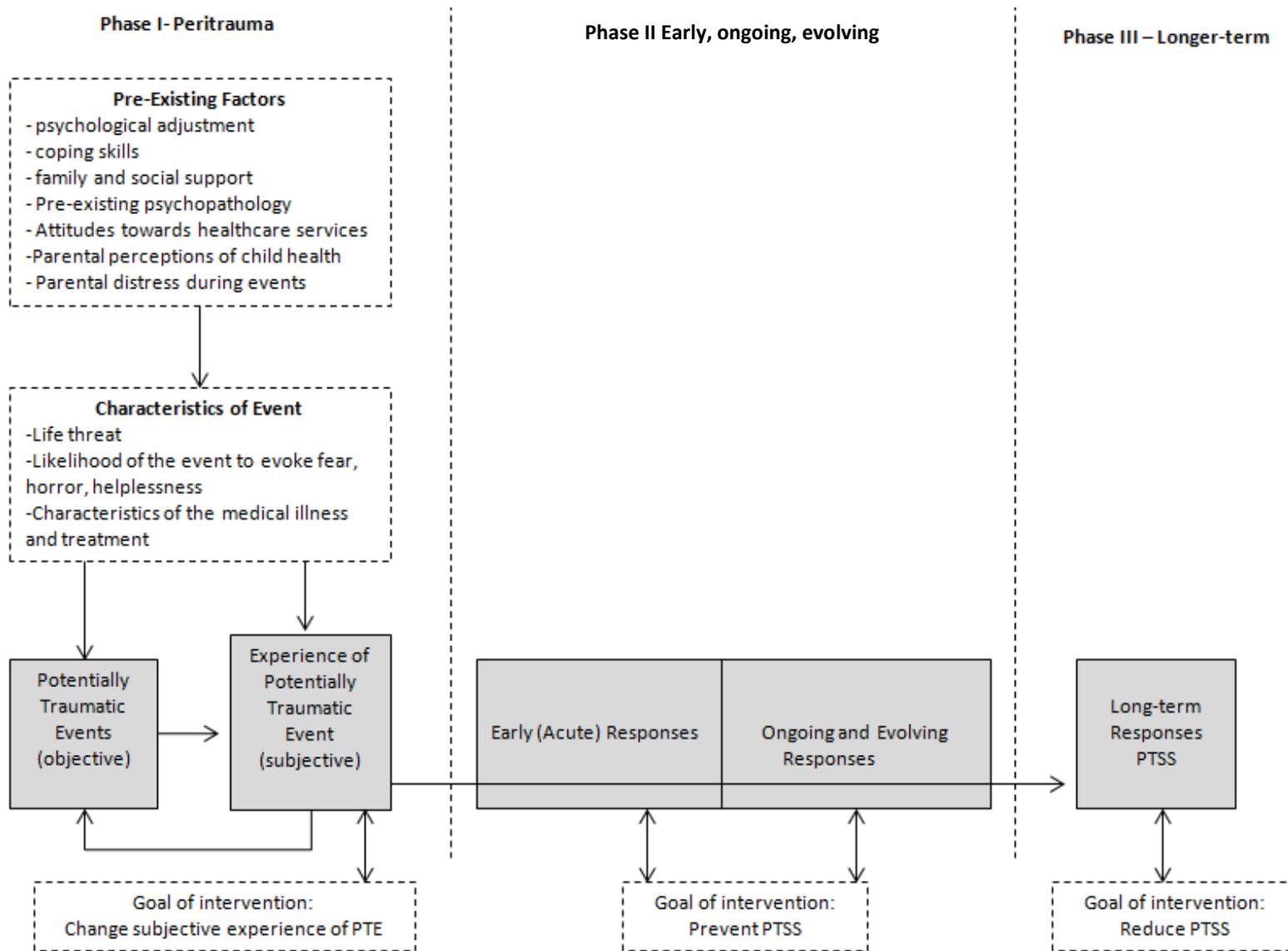


Figure 1.0.1 Paediatric Medical Traumatic Stress Model (Kazak et al., 2006)

1.6 Paediatric Medical Traumatic Stress

Kazak et al. (2006) proposed a three phase integrative developmental PMTS model to account for the physiological, psychological and behavioural reactions in children and families to medical procedures, pain, injury and illness. PMTS reactions were thought to include a continuum of traumatic stress symptoms of: physical arousal, re-experiencing and avoidance. The PMTS model emphasises the subjective experience of potentially traumatic events in determining whether a particular event is perceived as traumatic. Pre-existing factors such as: psychological adjustment, coping skills, parental distress, family and social support, and parents' general attitudes to health care services and perceptions of the child's health are proposed to play a role in this process (see Figure 1.1). These factors are thought to operate in conjunction with objective characteristics of the potentially traumatic event or illness such as: acuity, onset, length of exposure, intensity, threat of reoccurrence, complications, obviousness, number of family members directly involved and degree of human agency or responsibility (Kazak et al, 2006).

Phase one "Peritrauma" consists of the potentially traumatic event and its immediate aftermath. Phase two "Early, ongoing and evolving" describes traumatic stress responses to the sequelae of the traumatic event and medical treatment. Finally phase three "Longer-term", is the period after the resolution of immediate threats/ treatments when longer term traumatic responses occur. These phases are dependent on the nature of the medical condition and potentially traumatic events can be conceptualised as recurrent.

The model allows consideration of both the commonalities in the experience of potentially traumatic events across medical illnesses, and the disease or treatment specific factors which might influence whether and how PTMS is experienced by parents of

children with different conditions. The model can be applied to understand the potential pre-existing factors that may be involved in the experience of traumatic stress symptoms in parents raising a child with CHD. Whilst the model outlines potential factors which may contribute to an event being perceived as traumatic it does not describe the specific mechanisms by which this might occur. Any such mechanisms is likely to be highly personal and best described by a psychological formulation which considers the individual's personal pre-existing factors, the actual and perceived characteristics of the event highlighted in the model to give explanation to the experience of PMTS. As the model was developed before the DSM-5 diagnostic criteria was introduced it does not explicitly account for negative changes in cognition and mood. However, as the model does not describe an easily generalisable mechanisms for specific symptoms adding negative changes in mood and cognition to the symptoms of PTSS is less problematic. Something else not accounted for in the model is the experience of posttraumatic growth in parents of children with medical illnesses. The importance of recognising the possibility of the experience of positive changes alongside the experience of PMTS has been recognised by proponents of the PMTS model (Kassam-Adams, 2006).

1.7 Posttraumatic growth

Posttraumatic growth (PTG) has been thought of as positive changes following the struggle with traumatic events and adversity (Calhoun & Tedeschi, 2001). This construct has been known by a number of different names in the literature such as: perceived benefits, stress related growth, adversarial growth, positive psychological changes, thriving and adversity activated development (Linley & Joseph, 2004; Tedeschi & Calhoun, 2004). Joseph & Linley (2008) make the distinction between psychological wellbeing and subjective wellbeing. They assert that psychological wellbeing is about the

higher order “engagement with the existential challenges of life” (p11); whilst subjective wellbeing is about the experience of the subjective states such as distress. So that PTG indicates an increase in psychological wellbeing rather than subjective wellbeing.

Additionally it has been suggested that rather than indicative of a mental disorder PTSD symptoms such as re-experiencing, hyperarousal and avoidance can be considered normal subjective states experienced in the response to traumatic events indicating the need for cognitive-emotional processing of trauma-related information (Joseph & Williams, 2005) suggest. Thus both traumatic stress symptoms and PTG can be experienced concomitantly. A meta-analysis confirmed an overall relationship between traumatic stress symptoms and PTG, moderated by trauma type and age (Shakespeare-Finch & Lurie-Beck, 2014). The results of the meta-analysis were found to support both a linear and a curvilinear relationship. Whilst support for a curvilinear relationship was found to offer a better explanation statistically, the effect sizes of both solutions were similar (Shakespeare-Finch & Lurie-Beck, 2014).

There are a number of different models of PTG and the underlying theoretical structure of the construct remains unclear (Joseph & Linley 2008). Several different instruments have been used to measure PTG and have provided uni- and multi-dimensional factor structures for PTG (Linley & Joseph, 2004). Principle components analysis of three of these instruments Posttraumatic Growth Inventory (Tedeschi & Calhoun, 1995), Perceived Benefits Scales (McMillen & Fisher, 1998) and the Thriving Scale (Abraído-Lanza, Guier, & Colón, 1998) suggest that whilst PTG is uni-dimensional it has three broad components: changes in life philosophy, changes in perception of self, and changes in relationships with others (Joseph, Linley & Harris, 2004). This is also consistent with Calhoun & Tedeschi’s (2001) original conceptualisations of PTG.

The evidence around the beneficial significance of PTG is mixed. In their reviews on PTG both Linley & Joseph (2004) and Zoellner & Maercker (2006) conclude that whilst the cross-sectional evidence of a relationship between PTG and PTSD, and other indicators of psychological distress, is inconclusive, longitudinal evidence suggests that PTG is maintained over time and predicts lower levels of future distress. Some critics have challenged the phenomena of PTG as a defensive and illusory mechanism rather than a real experience of growth (Wortman, 2004). Zoellner & Maercker (2006) have taken this into account and combined both the “functional” and “illusory” sides to PTG in their two-component Janus faced model by asserting that PTG can be both a process and an outcome. Nonetheless, this criticism highlights deeper issues with the measurement of subjective states of psychological wellbeing using self-report tools and the debatable nature of the frequent assumption of absence of psychopathology as an indicator of psychological wellbeing.

1.7.1 Posttraumatic Growth in parents of children with disabilities and health problems

PTG has been investigated in: parents of children with illnesses such as diabetes and cancer (Hungerbuehler, Vollrath, & Landolt, 2011), parents of children who have survived stem cell transplantation (Forinder & Norberg, 2014); parents of children admitted to a paediatric intensive care unit (Colville & Cream, 2009) and parents of children undergoing corrective surgery for congenital birth defects (Li, Cao, Cao, Wang, & Cui, 2012). Whilst these studies have investigated different medical conditions and have used different time frames; ranging from directly after surgery (Li et al. 2012) to three years post diagnosis (Hungerbuehler et al., 2011), they all used the Posttraumatic Growth Inventory in a quantitative approach to identify the presence of perceived changes

in parents. In addition they all reported a significant level of PTG reported by parents, with prevalence ranging from 54% (Li et al., 2012) to 88% (Colville & Cream, 2009). This would suggest that PTG is experienced by parents of children with physical health problems and thus could be a relevant concept in understanding their experience.

However, whether what was measured by these studies truly captures the experience of PTG of these parents is debateable because, like much research from a nomothetic stance, the scale used was developed by researchers based on a theoretical conceptualisation of PTG. Tedeschi & Calhoun (1996) report the items on the inventory were developed based on a literature review of the perceived benefits following trauma. Furthermore the scale was then developed on a large sample of university students who reported to have experienced a significant negative life event. Thus it is possible that the experience of PTG in parents of children with health problems is qualitatively different and not completely captured by the questionnaire used.

PTG has also been investigated using qualitative methods in parents of children with disabilities (Scorgie & Sobsey, 2000). Through semi-structured interviews with 15 parents, they found that parents reported positive changes as a consequence of raising a child with a disability in three main areas: personal growth, improved relationships with others, and changes in philosophical or spiritual values (Scorgie & Sobsey, 2000). How these themes were expressed in the subthemes reported in the study richly described the experience of PTG for parents of children with disabilities but also maps onto tripartite conceptualisations in the literature.

1.7.2 Posttraumatic Growth in Parents of children with CHD

A systematic literature search (see Appendix B for search strategy) found a one study describing thematic analysis of 20 semi-structured interviews with adolescents and young adults with CHD and their parents in Australia. The aim of the pilot study was to determine whether the experiences reported fit an adversity-activated development (AAD) model of PTG (Kaisar, Strodl, Schweitzer, & Radford, 2012).

1.7.2.1 Adversity-activated development model of PTG

This model conceptualises responses to traumatic events as: positive, negative and neutral (Papadopoulos, 2007). The negative effects of trauma are divided into three degrees of increasing severity: ordinary human suffering, distressing psychological reaction and psychiatric disorder. PTSD falls into the last category. Thus not all negative responses to trauma are viewed as pathological. Neutral responses were constructed as resilience; an ability to “return to normal”. Positive responses to trauma in the model were termed adversity-activated development (ADD). Whilst AAD has been thought of as a term interchangeable with PTG (Joseph, et al., 2004). Papadopoulos (2007) proposed the construct differed from PTG in three ways. Firstly the name of the construct centres facing adversity rather than traumatic experience. Secondly the model does not assume a linear sequence in comparison to the implied meaning in *posttraumatic* growth. Positive effects were thought to be able to be experienced during adversity especially as adversity may continue. This proposition is supported by findings from meta-analysis (Shakespeare-Finch & Lurie-Beck, 2014). Thirdly the model exchanges the term “growth” for “development” which was considered a neutral word that encompasses a wider range of experience (Papadopoulos, 2007).

1.7.2.2 Application of the ADD model to the experiences of parents of children with CHD

The study by Kaisar et al (2012) found parents of young adults and adolescents with CHD described positive development such as gratitude and the acquisition of new coping skills. Parents also described both negative effects of their child's difficulties and resisting negative changes or resilience. It was concluded that the ADD model was relevant to this population as it acknowledged a wide range of responses to adversity (Kaisar et al., 2012). Whilst the findings of this pilot study are interesting the report of the study lacked clear description of the sampling strategy, inclusion criteria and sample demographics, challenging the transferability and dependability of the qualitative findings. Additionally the method of analysis was alluded to but not clearly described and there was little transparency in how the themes were generated or acknowledgement of how the researchers' own position might have influenced the analytic process, limiting the confirmability of the findings. Direct quotes were used to evidence the themes enhancing the credibility of the findings.

Whether these parents' experiences of ADD are similar to the experiences of parents with younger children is important as diagnosis and treatment for CHD most often occurs in the early months and years. Additionally (Kaisar et al., 2012) did not explore traumatic stress symptoms and the relationship of these symptoms to PTG in parents of children with CHD. PTG may be more apparent during the early years if it is also a coping strategy. Thus coping has been linked to both PMTS and PTG and would be of interest in this population.

1.8 Coping

1.8.1 Transactional stress and coping model

Coping has been defined as cognitive and behavioural efforts to manage the internal and external demands of situations that are appraised as outstripping an individual's resources (Lazarus, & Folkman, 1984). A review found that several studies of families with a child with CHD have used the transactional stress and coping model (Lazarus, & Folkman, 1984) to conceptualise the process of coping (Jackson, Frydenberg, Liang, Higgins, & Murphy, 2015). This model proposes that how a potential stressor is experienced by an individual depends on the interaction between factors which include: the environment, the individual's beliefs, personality, resources and repertoire of coping strategies (Lazarus, & Folkman, 1984). These factors are proposed to influence both an individual's primary appraisal of the significance and threat posed by the stressor and the individual's second appraisal of what can be done and how they will cope with the stressor. Lazarus and Folkman (1984) make a distinction between two types of coping behaviours. Problem-focussed coping, also known as instrumental coping, are those behaviours that approach the problem such as information seeking. Whereas emotion focussed coping, also known as palliative coping, are behaviours that deal with the emotional response to the problem, for example avoidance.

1.8.2 Coping, PTSD and PTG

Coping behaviours can be a risk factor for developing PTSD. The DSM-5 give "self-blaming or fatalistic coping strategies" p277 as examples of environmental pre-trauma risk factors and "inappropriate coping strategies" p278 as a posttraumatic risk factor for developing PTSD (APA, 2013). What inappropriate coping strategies are is not defined in this context. In addition to debates about whether PTG is a coping strategy or

outcome different coping behaviours have also been associated with PTG. In their systematic review, problem focused coping, acceptance, positive reinterpretation, religious coping, and emotion focussed coping were found to be associated with PTG (Linley & Joseph, 2004). Coping skills also feature in the PMTS model as a pre-existing factor proposed to influence whether potentially traumatic events are perceived as traumatic.

1.8.3 Coping in parents of children with CHD

A number of studies have examined associations between coping strategies, parental psychological distress and other variables in parents of children with CHD. These will be discussed together.

1.8.3.1 Coping and psychological distress in parents of children with CHD

Rychik et al. (2013) examined PTSD, anxiety, depression and coping in pregnant women whose unborn child had been diagnosed with CHD. They found that higher use of denial coping strategies was significant associated with higher traumatic stress, anxiety and depression. Higher use of acceptance coping strategies was significantly associated with lower depression and anxiety and higher use of positive reinterpretation and growth coping strategies was significantly associated with lower anxiety. In mothers of children with CHD it was found that daily hassles and palliative coping together accounted for 37.5% of the variance in “maternal adjustment” measured using the Brief Symptom Inventory (BSI) after the effects of CHD severity and maternal education were controlled for (Davis, Brown, Bakeman, & Campbell, 1998). Similarly it was reported that the use of behavioural disengagement, poorer understanding of their child’s CHD, and less family cohesion predicted poorer maternal mental health (Doherty et al. 2009). While for fathers

worry and the use of alcohol to cope predicted poorer mental health (Doherty et al. 2009). Taken together these findings might suggest that certain types of coping strategies are associated with poorer parental mental health. In contrast Spijkerboer et al. (2007) reported that parents of children with CHD demonstrated better psychological wellbeing than published reference groups. Parents of children with CHD reported less use of coping behaviours in the expression of emotion and reassuring thoughts scales; mothers reported less use of coping behaviours in the palliative scale and fathers reported less use of behaviours in the passive coping scale on the Utrecht Coping List (Spijkerboer et al., 2007). However the 60 item COPE used by (Doherty et al., 2009) and (Rychik et al., 2013) has 15 coping subscales whilst the 65-item ways of coping scale used by Davis et al. (1998) has only two, and the 44 item Utrecht Coping List used by Spijkerboer et al. (2007) {Spijkerboer, #372} has 7 subscales of coping behaviour, giving different coping profiles, making direct comparison of the findings more difficult.

The differences in these findings could also be due to differences in age of the children of parents in these studies. Whilst the children of parents in the Davis, et al., (1998) study were of wide ranging age, the children of parents in the Spijkerboer et al. (2007) study were at least 2 years older than the children in both the other studies. In the sample in the Spijkerboer et al. (2007) study surgery had been completed at least seven and a half years earlier and their heart conditions may have also been less severe than the heart conditions of children in the other studies. Using the severity ranking list used in the Davis et al. (1998) study, where higher numbers indicate more severe conditions, 70% of the children in the Spijkerboer et al. (2007) study were rated under a 6 compared to 40% in the Davis et al. (1998) study. Prenatally diagnosed CHD as in the Rychick et al, (2013) sample is also likely to be more severe than postnatally diagnosed CHD.

The three studies used different screening tools for psychiatric problems and disorders to assess psychological functioning: BSI (Davis et al., 1998; Doherty et al., 2009), General Health Questionnaire (Spijkerboer et al., 2007) and the Beck Depression Inventory and the State-Trait Anxiety Index (Rychik et al., 2013) making it difficult to compare the findings from the studies. In the Davis et al. (1998) study “Maternal adjustment” was measured using the BSI, suggesting that successful adjustment can be measured by the absence of symptoms of psychological distress. This assumption could be challenged as adjustment in raising a child with CHD could also be considered more than an absence of distress symptoms. This distinction echoes the distinction between subjective wellbeing and psychological wellbeing in PTG described previously (see section 1.6). Models of PTG would allow for the side by side experience of symptoms of distress, and positive growth or adjustment to traumatic experiences.

Much of the research in this area has focussed on determining adaptive and maladaptive coping patterns associated with better psychological functioning, as indicated by an absence of psychological distress. However the findings are mixed and no reliable patterns of relationship between coping and patterns and psychological functioning have been established. Bonanno (2013) suggests whilst it might be tempting to want to conclude that certain coping strategies such as “active” problem focussed strategies are helpful for adaptive adjustment one type of coping strategy is unlikely to be effective in all situations. This position recognises that, depending on the contextual factors of the situation other strategies may be more adaptive. Bonanno (2013) goes on to propose that adaptive psychological adjustment might be better facilitated through a flexible use of a range of coping strategies.

1.8.4 Coping Flexibility

Coping flexibility has been broadly defined as an individual's ability to adaptively use a range of coping strategies to facilitate adjustment to an ever-changing environment and has been operationalised in different ways (Cheng, Lau, & Chan, 2014). In a meta-analysis of the relationship between psychological adjustment and coping flexibility, operationalisations of coping flexibility based on strategy-situation fit and perceived ability conceptualisations demonstrated larger effect sizes compared to other conceptualisations of coping flexibility (Cheng et al, 2014). Perceived ability conceptualisations of coping flexibility have been measured using the Perceived Ability to Cope with Trauma Scale (PACT) (Bonanno, Pat-Horenczyk, & Noll, 2011) which specifically asks participants to report their perceived coping ability in response to a hypothetical potentially traumatic future event and maybe of relevance in understanding differences in the experiences of parents of children with CHD.

1.9 Rationale for current study

Previous studies have found differing levels of PTSD symptoms in parents of children with CHD, at different critical points, using different measures based on previous versions of the DSM criteria. In addition the sequelae of traumatic events are not always experienced as entirely negative and meta-analysis has shown that PTG can be experienced concomitantly (Shakespeare-Finch & Lurie-Beck, 2014). Whilst PTG has been investigated in parents of children with other physical health conditions there is little research on this construct in parents of children with CHD, particularly with younger children, currently or recently facing traumatic experiences.

The PMTS model suggests that both pre-existing characteristics such as coping and objective factors related to the child's condition are likely to be involved in whether a potentially traumatic event is perceived as traumatic, thus provoking traumatic stress and growth reactions. Coping is one variable of interest in both PMTS and PTG and in conceptualisations of PTG as a coping process or outcome of encountering adversity. Studies have found that different coping strategies have been found to be associated with psychological distress in parents of children with CHD. However this conceptualisation of coping does not consider whether the choice of coping strategy is adaptive or responsive to the circumstances and coping flexibility has not been assessed in this population before. A literature search combining PTSD, PTG and coping confirmed that this combination of variables has not been evaluated in parents of children with CHD in a single study before.

Finally much of the qualitative research into the experience of parents raising a child with CHD has focussed on understanding the process of parenting, the specific challenges and how they are managed. This study in contrast aimed to use a mixed methods approach to explore the perception and experience of traumatic stress symptoms, and PTG by parents raising a child with CHD in the UK.

1.9.1 Aims & research questions

The study's aims were to explore the experience of parents of a child with CHD as related to the constructs of traumatic stress symptoms and posttraumatic growth.

The research questions of the study were:

- 1) Do parents report traumatic stress symptoms related to their experience of raising a child with CHD?
- 2) Do parents report posttraumatic growth related to their experience of raising a child with CHD?
- 3) How do parents understand their experiences in raising a child with CHD?

2. Methodology

2.1 Epistemology ontology, paradigms and current study methodology

A researcher's epistemological assumptions are the ideas held about knowledge; what can be known and what it means to know something. Linked to this are ontological assumptions or the ideas held about the nature of being. The epistemological and ontological stances of a research study define the boundaries around what is considered data and how data can be understood and interpreted to arrive at knowledge (Hall, 2013). Epistemology also influences the assumptions which underpin judgements about how the quality of knowledge should be made. Related to epistemology and ontology is the term "paradigm" first coined by Kuhn (1970). The term "paradigm" has been widely used with many different interpretations of its meaning and as many ideas as to how the construct of paradigms should be used in relation to research. Morgan (2007) highlighted four meanings of the term paradigm as: worldview, epistemological stance, shared beliefs amongst researchers, and model examples of research. Two dominant and competing paradigms, in Morgan's (2007) second meaning of the term, have been described in psychological research; positivism and constructivism.

2.1.1 Positivism

Research within psychology has been described as historically set within a positivist paradigm. This realist position asserts that an external reality exists and can be measured. Research from a positivist paradigm, aims to discover truth and generalisable laws through objective means (Robson, 2011). Thus this paradigm is associated with empiricism and quantitative research methods which emphasise concepts such as: external and internal validity, reliability, generalisability, objectivity and the reduction of bias in assessing the quality of research from this paradigm.

2.1.2 Constructivism

A constructivist paradigm, in contrast to a positivist paradigm, takes a relativist position in asserting that reality is subjective and meaning is constructed through interactions. Robson (2011) suggests research from this paradigm is idiographic and aims to understand multiple perspectives. Additionally the knowledge produced by research from this paradigm would be thought to be situated within a specific context and co-constructed with participants. This paradigm is associated with qualitative research methods. Constructs such as credibility or the believability of the findings from the perspective of research participants, transferability or the degree to which findings can be generalised to other contexts, confirmability or the degree to which results can be corroborated by others, dependability and transparency are relevant in assessing the quality of research from this paradigm (Lincoln & Guba, 1985).

The incompatibility of these contrasting epistemological stances gave rise to the “paradigm wars”, in which these paradigms were pitted against each other (Bryman, 2008). Mixed methods research, where quantitative and qualitative methods are combined within a single study, evolved in this context as a way forward. Some described mixed methods research as a “third research paradigm” which aimed to bridge the gap between the paradigms and combine the strengths of one research method to complement the weaknesses of the other (Johnson & Onwuegbuzie, 2004; Morgan, 2007). This combining of research methods from different paradigms has led to ongoing debates about the complementarity or incommensurability of paradigms (Masse, 2000), and the appropriate epistemological stance for mixed methods research (Johnson, Onwuegbuzie, & Turner, 2007)

2.1.3 Critical realism

A critical realist epistemological stance has been constructed as an alternative epistemological and ontological position between positivist and constructionist paradigms (Sayer, 2000). It has been described as a combination of realist ontology and constructivist epistemology (Maxwell & Mittapalli, 2010). Critical realism has a realist ontology in that it assumes that an external reality exists, independent of our knowledge and understanding of what exists. However critical realism also asserts that reality can only be known through our partial and situated perspectives, including the discourses available, and thus can be considered fallible and open to change (Sayer, 2000). Critical realism allows for the side by side coexistence of different perspectives and has been associated with a range of research methods including mixed methods research.

Critical realism takes a distinctive view of causality, distinctive from perspectives underpinned by regularity view of causation which propose that regularity in a relationship is a precursor to inferences of causation, most typified in Hume's term "constant conjunction" (Maxwell, 2004). Critical realism has been described as taking a process oriented approach in which objects and social relations are thought to have causal powers which may or may not produce regularities (Sayer, 1992). Critical realism also considers social systems to be open and changeable, which makes finding regularities and nomothetic laws difficult. However critical realism focuses on necessity and contingency in causation rather than regularity and prediction (Sayer, 2000). Thus associations are possibilities in a system rather than indicative of certain causalities. Furthermore mental experiences, which are thought of as existing in reality, can also be considered causes.

2.1.4 Criticisms of paradigms

The conceptualisation of paradigms has been criticised as an arbitrary bringing together of different epistemological and ontological assumptions which give the impression that paradigms must be bought in a set menu (Biesta, 2010). Biesta (2010) argues that this use of paradigms can preclude thoughtful discussions about the primary epistemological assumptions that underpin research. How these are conceptualised will influence the research questions that are asked within a study and the methodology employed. Indeed some have claimed that understanding epistemology and ontology are essential in developing the methodological decisions in the research (Guba & Lincoln, 1994). Other positions such as pragmatism take a lighter position on the necessity of a connection between epistemology and research methodology in an attempt to sidestep the issue of the incommensurability of paradigms.

2.1.5 Pragmatism

Pragmatism has been described by some as the epistemological stance most suitable for mixed methods research (Johnson & Onwuegbuzie, 2004; Morgan, 2007). However Biesta (2010) suggests that whilst pragmatism, can offer some perspectives on issues in mixed methods research, it cannot provide a philosophical basis for mixed methods research. For this reason this study did not take a pragmatic position whilst it did take a view of paradigms as tools to guide the decision about the epistemological stance of the study.

2.1.6 Current study aims and epistemological position

The current research aimed to explore the impact of the difficult experiences in raising a child with CHD for parents with respect to the constructs of traumatic stress

symptoms and PTG. The epistemological position most closely aligned with this study is a critical realist stance which would view the phenomena of: traumatic stress symptoms and PTG as existing in reality independent of our understanding of them and the discourses through which we understand them. These discourses would be assumed to be imperfect and more or less helpful. Thus a critical realist position would support the use of quantitative methods to assess the phenomena, as one way through which the phenomena can be understood. At the same time a critical realist stance would also emphasise the subjective nature of experience and the individual's understanding and the meanings made of experience. Thus this position would also support the concurrent use of qualitative methods to understand the individual's experience and the shared construction of this within a group. A critical realist stance would also accept that observer bias in qualitative research cannot be removed, and thus emphasise in keeping with Greenhalgh & Taylor's (1997) recommendation that researchers reflexively describe their positioning so that research findings can be interpreted accordingly.

2.2 Study setting

One of the challenges of research with parents of children with health conditions is participant recruitment. This can be for a number of reasons such as: low prevalence rates of the condition leading to a smaller pool of potential participants and additional barriers to participation such as time constraints related to participants' caring role for their child, for example: additional health related care tasks, burden of childcare, burden of healthcare costs. For these reasons traditional research methods are not well suited for this group. Newer internet facilitated research methods, such as internet based forums offer flexibility, convenience and consequently increased accessibility for participants. These methods also have advantages for researchers in the lower cost and accurate and

ready data format. These methods have been found to yield similar findings to traditional face to face methods (Kramish Campbell et al., 2001). One of the disadvantages of this data gathering tool is selection bias described as an effect of the “digital divide” (Kramish Campbell et al., 2001) where those who have access to the internet and those who might use forums on the internet tend to be younger and come from more affluent social groups. Nonetheless internet use in the UK continues to grow with nearly all households with children (99%) having the internet in 2016 (Office for national statistics (ONS, 2016). Additionally 82% of adults reported having used the internet every day or almost every day, with 63% of adult internet users looking at social media sites at least weekly (ONS, 2016).

Online discussion groups can be considered a form of bulletin board focus group, where a set number of individuals participate in asynchronous electronic discussion (Krueger & Casey, 2009). It was hoped that asynchronous online discussion group would allow potentially geographically distant parents, who were likely to have caring duties, to respond to the questions and engage in asynchronous conversation with other parents at a time and location of their choosing. Thus this method would allow parents to form a virtual focus group that cuts across time and geographic location. The asynchronous element could also allow parents to reflect in more depth on their responses as there is less time pressure to respond and less of a demand on parents’ ability to type quickly (Krueger & Casey, 2009).

Participants were recruited through the Children’s Heart Federation (CHF), the leading UK registered children’s heart charity. The charity provides: information and support to parents, advice to policy makers and service providers, support for research

projects and support for campaigns for improvements to service provision for children and families with CHD. The CHF also had experience of collaborating with researchers and using a Facebook; a social media site, as a medium for data collection in a previous study (Brown et al., 2016). Brown et al. (2016) elicited parents' experiences of caring for a baby with CHD at home after surgery, through questions posted on a large online forum on Facebook. Of the 91 participants given access to the closed forum, 73 parents participated, responding to between 1 and 5 questions. However, the large number of participants in a single forum may have discouraged some parents from participating and sharing more personal or difficult aspects of their experience, or promoted social loafing (Shiue, Chiu, & Chang, 2010).

2.3 Study design

A priority sequence approach (Morgan, 1998; 2014) was used in conceptualising the mixed methods design. A preliminary cross-sectional quantitative method was conducted to facilitate selection and description of participants and to answer research questions asking about levels of PTSD and PTG. This was followed by a core qualitative method. Interested parents were invited to participate in discussion groups based. The aim of the qualitative element of the study was to explore how parents construct their experience of the impact of the difficult experiences in raising a child with CHD with respect to constructions of PTSD and PTG. The online discussion groups were facilitated on Facebook by the researcher using a study account. To protect confidentiality the Facebook groups were closed groups (secret group on Facebook) so only group members and the researchers facilitating the forum were able to view the group and the comments posted by participants. Only the primary researcher was able to add participants to the group.

2.3.1 Online focus group size

It has been suggested that the optimal focus group size is 5-8 participants (Krueger & Casey, 2009). Where participants have a lot to share, or where the group might also act as a platform to share tips and information, fewer participants have been advised (Krueger & Casey, 2009). As the study was conducted online using an asynchronous format it was anticipated that participants would be able to share more information, in more depth and of a more reflective nature than face-to-face focus groups might ordinarily permit. Thus the study aimed to recruit a maximum of eight participants per group.

2.4 Method of recruitment

The CHF facilitated an information page about the study (Appendix C) on their research webpage which contained a link to the quantitative survey. The CHF posted a brief Facebook advertisement (see Appendix D for advertisement wording) which contained a link to the information page. The CHF also tweeted a link to the study information page to their twitter network once weekly for a period of four weeks.

2.4.1 Inclusion criteria

Parents were eligible for the study if they were aged 18 and over, had a child diagnosed with a congenital heart disease and were able to communicate using written English.

2.4.2 Exclusion criteria

Parents were excluded from the study if their child had additional developmental syndromes in addition to CHD Parents were also excluded from the qualitative element of

the research study if they could not access the internet regularly or were unwilling to open or use a Facebook account for the purposes of the study.

2.5 Materials

Three standardised self-report questionnaires and a demographic data collection tool were used in the quantitative element of the study. These were administered as a 5 page online survey using Qualtrics software (2015).

2.5.1 Demographic data collection tool

The following demographic information about the parent participants and their child was collected to contextualise the study findings and inform sample description: parent age, parent gender and parent ethnicity, the child's congenital heart condition, time since the child's last treatment and the presence of additional developmental disorders. Finally the number of other children in the family and family structure were also collected (see Appendix E).

2.5.2 Measure of Coping Flexibility - Perceived Ability to Cope with Trauma Scale (PACT)

This 20 item self-report measure assesses respondents' beliefs about their ability to employ different ways of coping when facing a potentially traumatic event (Bonanno, et al., 2011). Thus the scale measured perceived ability. Respondents were asked to rate on a 7 point Likert scale how able they thought they would be to use the behaviour or strategy if they had to confront a potentially traumatic event (see Appendix F). The scale consists of two subscales: trauma focused and forward focussed coping. A mean score was calculated for each scale. Coping flexibility was calculated from the mean scores on the subscales using the formula described in Burton et al., (2012):

$$\text{Coping Flexibility} = \frac{(2S+1)}{(S+L+2)}$$

Where S is the smaller mean score and L is the larger mean score. Coping flexibility scores ranged from 0–1 with higher scores indicating high and equal use of both trauma focussed coping and forward focused coping strategies and thus more flexible coping.

The internal reliability of the trauma focussed scale was reported as ($\alpha= 0.91$) and the forward focussed scale ($\alpha= 0.79$) (Bonanno et al., 2011). The trauma focussed and forward focussed scales were found to have a moderate associations with ego-resiliency ($r= 0.30$) and ($r= 0.36$) respectively measured using the using the ER80 scale (Bonanno et al., 2011). Neither the trauma nor forward focussed scales were associated with social desirability measured using the Marlowe-Crown social desirability scale ($r= 0.01$) and ($r= 0.04$) respectively (Bonanno et al., 2011).

The PACT has been used with bereaved adults (Burton et al., 2012). Conjugally bereaved participants suffering from complex grief had significantly lower coping flexibility (0.79) than both the non-bereaved (0.84) and the asymptomatic conjugally bereaved (0.84) comparison groups. A meta-analysis found that coping flexibility had a moderate sized association ($r= 0.32$) with psychological adjustment assessed with measures of well-being, quality of life, psychological distress, and anxiety and depression (Cheng, et al., 2014).

2.5.3 Measure of Traumatic Stress Symptoms - PTSD Checklist for DSM-5 (PCL-5)

The PCL-5 is a 20 item self report measure which assesses the presence of symptoms of PTSD according to the DSM-5 criteria. The scale assesses the four DSM -5 clusters of symptoms: intrusions (items 1-5), avoidance (items 6-7), negative mood and cognitions (items 8-14), and arousal and reactivity (items 15-20). Respondents rate on a 5 point Likert scale how much they had been bothered by the symptom in the last month (0 = Not at all, 4= extremely). (see Appendix G). The scale gives a total symptom severity score, obtained by summing all item scores, which ranges from 0 – 80, with higher scores indicating greater pathology (Weathers et al., 2013). Symptom cluster scores can be obtained by summing the scores for items within the cluster. In a large sample of university students who self identified as having experienced a “very stressful life event” the PCL-5 was found to have good: internal reliability ($\alpha= 0.94$), test-retest reliability ($r= 0.82$) convergent validity against the Posttraumatic Diagnostic Scale ($r=0.85$) and the PCL-Specific ($r=0.85$) and discriminant validity against depression ($r = 0.6$) and anxiety ($r = 0.4$) (Blevins, Weathers, Davis, Witte, & Domino, 2015). As a relatively new measure a clinical cut point score of 33 has be recommended for use until further validation studies have been completed (Weathers et al., 2013).

2.5.4 Measure of Posttraumatic Growth - The Psychological Well-Being – Posttraumatic changes questionnaire (PWB-PTCQ)

The PWB-PTCQ is an 18 item self-report measure of posttraumatic changes. Respondents were asked to rate on a 5 point Likert scale the degree of change they perceived had occurred since the traumatic event (1= much less now, 5 = much more so now) (Regel & Joseph, 2010 cited in Joseph et al., 2012). Scores were summed to give a

total between 18 and 90. Unlike other measures of PTG the PWB-PTCQ also allows negative change to be captured. Scores above 54 suggest positive change with higher scores suggesting more positive change and lower scores indicating negative changes. The PWB-PTCQ has shown high internal reliability ($\alpha = 0.87$), good convergent validity with the posttraumatic growth inventory ($r = 0.56$), and good test-retest reliability ($r = 0.55$) and good incremental validity over and above other measures of PTG (Joseph et al, 2012). The instructions to the scale were adapted to specify the most distressing part of their child's diagnosis and treatment of CHD as the reference events of interest (see Appendix H).

2.5.5 Discussion group questions

Broad open discussion group questions were formulated to allow participants to bring the issues of importance to them (see Table 2.1). These questions were followed by suggestion prompts. Each question was discussed with the CHF prior to introducing to the groups to ensure the language used was clear and the meaning understandable.

Table 2.0.1 Discussion group questions and prompts

Week	Questions and Prompts
Ice breaker	<p>To help the group get to know each other could you respond to the following question: Where do you like to go with your family and why?</p>
Week 1	<p>Hello and welcome to the online discussion group Thanks for telling us about the family activities you enjoy. This week's question is: Please tell us a bit about your child's condition, his/her treatment so far and your experience of this.</p>
Week 2	<p>Hello again, Thank you for all your comments and likes this past week. Thanks for sharing with us some of the things you and your child have had to face, and continue to face. This week I want to ask you about the most distressing or disturbing aspects, times or situations for you.</p> <p>What was most distressing/ disturbing for you? Can you describe what it was or what was going on? What was it that made these aspects/ times/ situations most distressing or disturbing?</p> <p>I appreciate some people may find this a difficult topic so please say as much as feels comfortable. Also feel free to come back to it to add more comments later, to your own or others comments, if it is easier. Do please share if you feel comfortable. We are grateful for your participation with this question and appreciate that it may bring challenging memories and feelings to the fore and as a group we are grateful for your kind sharing of your experiences.</p>

Table 2.1 (continued)

Week	Questions and Prompts
Week 3	<p>Hello everyone, I just want to say thank you for all your time and contributions so far I realise that life can get really busy and it can be hard to find time, so thanks again. This week I'd like to ask you about how you got through those situations and difficult times.</p> <p>What helped you to get through and also what was not helpful? What would you have liked to be done differently by others? What would you have done differently yourself?</p>
Week 4	<p>Hello Everyone, Thanks for sharing what helped you get through and your honest thoughts about what was helpful and what was less helpful.</p> <p>Q4. This week I want to ask about your reactions and the impact on you. This could be at the time of the most distressing/ disturbing events or afterwards, whatever you have noticed.</p> <p>What reactions did you have and what has been the impact on you? What thoughts, images, feelings or sensations did you experience? What did you make of these experiences?</p> <p>Again some people may find this a difficult topic so please say as much as feels comfortable and please do let me know if it becomes too much.</p>
Week 5	<p>Hello Everyone, Thank you for sharing your thoughts and experiences on the topics so far. This is our penultimate week.</p> <p>Q5. This week I want to ask about the longer term impact on your life. Again this could be in relation to the most distressing events or it could be on the whole, whatever you have noticed.</p> <p>What are the differences, if any, in your life now? What has the impact been? What changes, if any, have you noticed within yourself or about you as a person? What changes, if any, have you noticed in your family or in the people around you?</p>

Table 2.1 (continued)

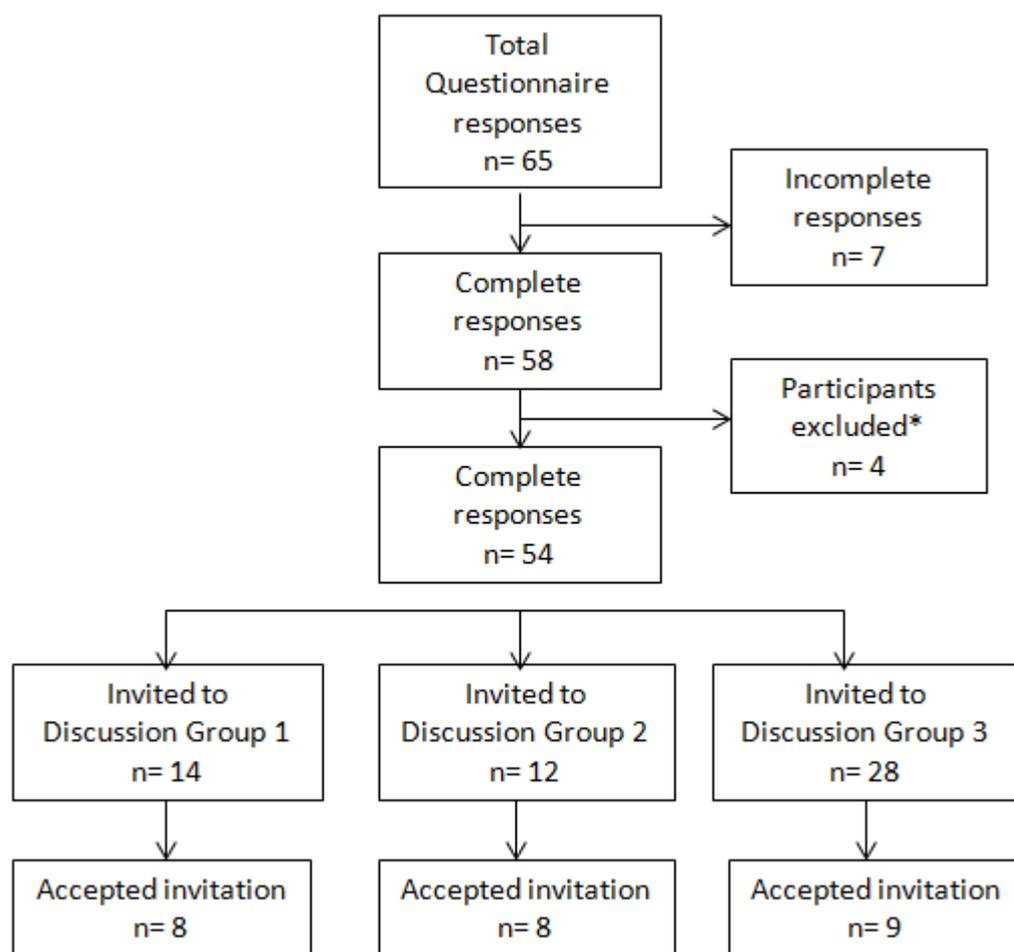
Week	Questions and Prompts
Week 6	<p data-bbox="475 309 1362 342">Hello Everyone,</p> <p data-bbox="475 347 1362 423">This is our last week. I wanted to thank you for sharing your stories and experiences with the group over the last few weeks.</p> <p data-bbox="475 427 1362 504">This week I wanted to ask about what it has been like for you to take part in the discussion groups.</p> <p data-bbox="475 551 1362 584">What, if anything, was helpful and what, if anything, was unhelpful?</p> <p data-bbox="475 589 1362 665">Is there anything else you think is relevant to the discussions that you have not been able to mention previously?</p> <p data-bbox="475 712 1362 828">For this week I am removing the CHF as observers as they would also like some confidential feedback on the materials and support they provide.</p> <p data-bbox="475 833 1362 949">What do you think the CHF could do differently to support parents and heart children? What do you think of the materials and support they provide?</p>

2.6 Research Procedure

2.6.1 Phase one: Quantitative survey

Access to the quantitative survey was through a link at the bottom of the information page about the study on the CHF website. The consent statements formed the first page of the survey (see Appendix I). Parents were required to indicate agreement with each consent statement before access to the questionnaires was granted. The last question of the consent statements asked participants if they were willing to participate in an online discussion and if so they entered their email addresses so they could be contacted at a later date with further information about this aspect of the study. The demographic questionnaire was then presented, followed by the PACT, PCL-5, and PWB-PTCQ. Each questionnaire was presented on one page and the questionnaires were presented in the same order for all participants. Participants could opt out by closing or navigating away from the questionnaire pages at any stage. The quantitative survey data

collection took place over three weeks during August 2015. Data was exported from the Qualtrics website into SPSS. A list of participant identification numbers and email addresses of participants interested in participating in the discussion groups was saved separately in a password protected document.



*Participants meeting exclusion criteria – additional development disorders

Figure 2.0.1 Questionnaire responses and online discussion group formation

2.6.2 Phase two: Qualitative online discussions

2.6.2.1 Group formation

All participants left a contact email address to be contacted for the discussion groups. After the quantitative survey phase ended, perceived coping flexibility was

calculated (see section 2.5.2) for those who submitted complete responses and met the inclusion criteria. Participants were purposefully selected to include parents with a range of coping flexibility across the sample. This increased the likelihood of variability of in the sample as coping has been theorised as a factor variable in the PMTS model (Kazak et al., 2006). To achieve this, participants were rank ordered according to their total perceived coping flexibility score. The eight participants with the highest and lowest coping flexibility scores were invited to join online discussion groups by email. Participants were sent instructions for gaining access to the group (Appendix J) and guidelines for using the forum which included considerations for keeping the forum secure (Appendix K). As participants responded to the invitation they were added to the discussion groups on Facebook. This procedure was repeated until both groups had eight participants. Fewer participants responded to the invitation to join group one so more invitations were sent to participants with low coping flexibility scores before the group was filled. The remaining participants were then invited to join group three.

2.6.2.2 Data collection

Three online focus groups were conducted over a six week period with a different discussion question presented each week. When the first participants were added to the group a welcome message and ice-breaker question was posed to participants so that participants could get to know each other and get used to posting comments in the group before the research questions were posed.

The researcher monitored the discussion group at least once a day. Where identifying information such as hospital names were posted the researcher reposted the message using Xs to preserve anonymity. Participants were informed that the CHF

research officer was a silent moderator in each of the groups. During the data collection period the researcher also posted three open prompts to the discussion groups, for example:

Hi everyone, Thanks for all your comments in the group so far. I notice fewer people have commented on this week's question. Is that because it's a difficult question? Please feel free to say that or whatever you want about the question. I'll be posting the next question tomorrow afternoon. Best wishes

Data was pseudonymised and exported into MAXQDA software (2015) for analysis and word-processed into transcripts without any other amendments to the text.

2.7 Analysis

2.7.1 Quantitative analysis

Demographic and questionnaire data were analysed using the Statistical Package for the Social Sciences (SPSS) version 19 (IBM Corp, 2010).

2.7.1.1 Data cleaning

Data was screened for missing values and visually inspected for outliers using box plots. Participants with missing data were excluded on a variable by variable basis.

2.7.1.2 Statistical Analyses

Data was visually inspected and the statistics for skewness and kurtosis for each variable were then examined by dividing the skewness and kurtosis statistic by the standard error (Field, 2009). If this figure was larger than ± 1.96 , $\alpha < 0.05$ level, it was taken to indicate the distribution differed significantly from a normal distribution and the mean and standard deviation were not used to describe the central tendency and

dispersion of scores. Descriptive statistics were calculated for each of the questionnaire measures for the whole sample and for contributing and non-contributing parents in the discussion groups.

2.7.2 Qualitative analysis

Braun & Clarke's (2006) thematic analysis was selected as the most appropriate method of analysis for this study because of its systematic and transparent approach. Unlike other analytical approaches such as Interpretive Phenomenological Analysis (Smith, 1996) where analytical procedures are based on particular epistemological assumptions, thematic analysis is a theoretically flexible method that can be used with research from a range of epistemological positions. Additionally unlike analytic approaches such as grounded theory (Charmaz, 2006) which has a set procedure with the aim of arriving at a bottom up derived theory, thematic analysis can be conducted in different ways depending on the researcher's aims and purpose (Braun & Clarke, 2006).

This study aimed to explore how parents construct the impact of the difficult experiences in raising a child with CHD. Of interest were the constructs of traumatic stress symptoms and PTG. From a critical realist stance, analysis at both the latent and semantic levels were of interest. One of the limitations of thematic analysis in constructing latent meaning and assumptions is that unlike discourse analysis thematic analysis is not a suitable method to examine the use of linguistic structure and language use (Braun & Clarke, 2013). However as an initial exploration this method was suitable and a bottom up approach was selected to achieve the study aims. Braun and Clark's (2006) six stages of thematic analysis were applied in this study as described below.

2.7.2.1 Familiarisation with the data

The discussion group data was read and re-read several times, to facilitate immersion in the data (Braun & Clarke, 2006). Initial reactions and ideas for coding were recorded by the researcher and were used in the next stage of analysis.

2.7.2.2 Generating initial codes

The transcript from each group was coded which entailed systematically dissecting and labelling elements of participant responses that referred to an idea or concept related to the research aims and questions. Both semantic or surface content meanings and latent meanings, or inferences about underlying assumptions or meanings were coded (see Appendix L for examples of the coding frame). The following recommendations from Braun and Clark (2006) were followed: data was coded for as many themes as possible, data extracts were coded inclusively to retain the context, and multiple codes were permitted for extracts.

Reflective memos were made on the transcript using MAXQDA (see Appendix M for an example). The software was then used to collate codes and manage the data. Responses to the first three questions of the first group coded were re-coded at the end of coding to ensure that coding remained consistent. After the data from each group had been coded the codes were reviewed by checking each coded item against the code. A list of codes was generated across the groups.

Multiple coding

A small section of one transcript was also coded independently by two additional coders to explore validity in the coding (Barbour, 2001). This exercise was not an attempt

to remove subjectivity in order to arrive at the same codes, as critical realist and constructivist epistemological stances would respect that different researchers will arrive at different interpretations, without it representing a threat to the soundness of the findings. Multiple coding was used in a reflective process to help refine and develop coding by presenting alternate interpretations and illuminate the researcher's lenses so that systematic, transparent and valid coding could take place (Barbour, 2001)

2.7.2.3 Developing themes

The codes were then manually sorted and developed into themes. This iterative process can be described as identifying patterns and links between the codes which related to the research questions. This involved: moving back and forward between codes and coded text, expanding, collapsing, and grouping codes to construct themes which made sense of the data in relation to the research question. This process was conducted several times.

2.7.2.4 Reviewing themes

Each theme was reviewed by checking the coded segments against the theme to develop and ensure coherence in the theme. Themes were constructed to minimise overlap between themes but retain connectedness. Notes were made on each theme during this process.

2.7.2.5 Defining and naming themes

Each theme was summarised, named and data extracts were identified which exemplified the theme. These were discussed in research supervision to facilitate the researcher's consideration of the dependability of the results of the analysis.

2.7.2.6 Producing the report

Finally the themes were written up in a narrative in the report, using direct participant quotations and retaining as much context as possible.

2.7.3 Integrating quantitative and qualitative analyses

The quantitative and qualitative methods were employed in a priority sequence mixed methods design. A final step involved integrating the results of the quantitative and qualitative analyses at the interpretation and reporting level (Fetters, Curry & Creswell, 2013) to facilitate understanding of the research findings. This involved presenting the different types of data together in an integrated discussion.

2.8 Methodological rigour

When establishing the methodological rigour of any research study, how a study conceptualises what can be known and how it can be known will guide decisions about what is considered sound and how a study should achieve this. This becomes more complicated in mixed methods studies and currently there is no consensus on how the quality and rigour of mixed method studies should be assessed (Brown, Elliott, Leatherdale, & Robertson-Wilson, 2015). As such, the criteria for assessing quantitative and qualitative methods tend to be applied to each component with special attention paid to the area of integration of mixed methods (Brown et al., 2015). These separate criteria will be considered here with respect to the current study.

2.8.1 Quantitative research

The accepted criteria for assessing the soundness of quantitative research are: internal validity; the degree to which an observed effect is attributable to the effect of the independent variable, external validity; the generalisability of the research findings,

reliability; the consistency of the research findings over time, and objectivity; the degree to which the research minimises bias. These criteria are based on a realist ontology and epistemology that assume true phenomena which are directly observable and measurable exist and research should aim to measure these phenomena as accurately as possible.

Internal validity is more relevant to experimental studies where variables are manipulated and thus less relevant to this study.

One way external validity can be addressed is by ensuring that sampling is representative and unbiased, with a random sampling being the gold standard. As the size of the base population of interest in this study is small and the sample was accessed through the CHF network, the quantitative findings from this sample are not intended to be generalised to wider populations. Quantitative measures were used to contextualise the qualitative element of the study and aid participant selection. Several factors were still considered in order to optimise the response rate and to minimise selection bias for the qualitative element of the study. This included advertising the study through the internet, Facebook and Twitter, reducing the burden of participation by limiting the questionnaires and demographic questions, and increasing ease of administration of the study by using online data collection platforms. In this study reliability was considered by using measures with good reported test-retest reliability and internal reliability where possible. The internal reliability of each measure within the current sample was also estimated as some measures had not been used before in this population and the gold standard is to use measures which have been validated in the population they are used (Prince, 2003).

2.8.2 Qualitative research

Despite the ongoing debate on how the quality of qualitative research should be assessed (Angen, 2000; Porter, 2007; Rolfe, 2006; 2007) Lincoln and Guba's (1985) four criteria, which broadly correspond to the criteria used to evaluate quantitative research, remain the gold standard for assessing the quality of qualitative research (Brown et al., 2015). These criteria are: credibility; the believability of the findings from the perspective of research participants, transferability; the degree to which findings can be transferred to other contexts, confirmability; the degree to which results can be corroborated by others and dependability; the degree to which changes in the context and how these affected research process are described.

The credibility of the study findings was considered in the following ways: use of multiple coders, discussions with research supervisors during analysis phase, staying close to the data during analysis, and using participant quotes to illustrate the themes. To allow the transferability of the findings; demographics, and questionnaire data were reported alongside description of the study setting to aid a thick description of the context of the study. The critical realist stance adopted in this study maintains a constructivist epistemology which also recognises that knowledge is partial, situated and thus fallible. These are challenges to the confirmability of the study findings. In recognition of this I kept reflective notes throughout the development and conduct of the study. I have also described my position and personal influences in section (4.6).

2.9 Ethical considerations

2.9.1 Ethical approval

Ethical approval from the University of Essex research ethics committee was sought and granted (Appendix N). As the study identified participants through a third sector organisation NHS REC approval was not required according to the “Do I need NHS Ethics Approval?” Health Research Authority Tool (2013). This was also confirmed by the Children’s Heart Federation.

2.9.2 Informed consent

Access to the survey questionnaires was through a link on the information page which explained: the study, the potential benefits and risks from taking part in the study, confidentiality, data protection, and their right to withdraw from the study at any stage and the right to withdraw their data from the study (see information page Appendix C). Participants were also given the contact details for the researcher and supervisors to provide the opportunity to ask questions prior to consenting to participate. The consent statements (see Appendix I) formed the first page of the survey questionnaires.

2.9.3 Participant vulnerability

The Code of Human Research Ethics published by the British Psychological Society (2010) was used to determine that research participants in this study were not classified as a vulnerable group. However the concept of vulnerability in this context was understood to relate to the issue of informed consent. In that, vulnerable groups require special consideration due to a diminished capacity to voluntarily make a decision free from coercion that protects their own interests. Some researchers have argued that whilst

survivors of traumatic events may not be categorised as a vulnerable group under this conceptualisation of vulnerability, they do require special consideration and care (Collogan, Tuma, Dolan-Sewell, Borja, & Fleischman, 2004). This was considered in the next section.

2.9.4 Participant welfare and managing the potential for distress

Participants scoring above clinical cut offs on the PCL-5 were contacted by email and informed that they might consider seeking medical advice if they were still troubled by the symptoms they reported. In total 11 (20.4%) participants were contacted in this way.

There was a small but potential risk that parents participating in discussion groups may have become distressed by discussing difficult experiences associated with their child's CHD. A review of the literature suggests that the likelihood of significant emotional harm to participants in trauma-focussed research studies is low (Newman, Risch, & Kassam-Adams, 2006). Furthermore the online, asynchronous nature of the study allowed participants greater control over: the physical environment, the timing of participation and in deciding whether to respond to the research question for the week, what and how much to say and whether to continue to participate in the study. Nonetheless, special considerations for managing the vulnerability of this potentially geographically diverse group of participants were still considered.

Participants were made aware of the potential risks of participation on the information page and during the consent procedure. Participants were reminded of the helpline operated by the CHF which they were able to access if they became distressed.

The researcher planned to direct participants to this support service in addition to advising them to seek medical attention. The online forum was checked by the researcher at least daily during the data collection period to monitor and moderate the forum. Participants were reminded to contact the researcher in the event of any difficulties and regular research supervision was scheduled during the data collection period to resolve any risk issues. Additionally midway and at the end of the discussion groups the researcher sent an individual message to each participant to check how they were, thank them for their participation and to remind participants of the researcher's contact details if they had concerns or required additional support.

At the end of the second week of data collection, one participant in the moderate perceived coping flexibility group dropped out of the study. In follow up correspondence the participant reported they had found participation distressing and they were already accessing additional psychological support and they did not want to discuss their experience in the discussion groups. This participant's comments in the discussion group were withdrawn from the study at their request. No other participants dropped out of the discussion groups. No other participants reported any concerns or ill-effects from taking part in the discussions at any point.

2.9.5 Data confidentiality

To protect participant identities all data was pseudonymised and securely stored. In keeping with University procedures following successful completion of the thesis and viva the original data will be destroyed; that is all posts to Facebook will be deleted and all participants removed from the group to allow the standard Facebook procedure of deletion of inactive groups without members to take place.

2.10 Dissemination

A written summary report based on this thesis will be disseminated to the Children's Heart Federation for publication on their website after their approval. This report will also be sent directly to participants who indicated their interest in the outcome of the study in the discussion groups. The findings from this study will be written up as a manuscript and submitted to an academic journal. The findings of the study may also be presented at conferences.

3. Results

This section presents descriptive statistics from the questionnaire data to understand whether parents, in this self selecting sample, report symptoms of PTSD and PTG. This is followed by the demographic information for the group discussion participants, and descriptions of the themes constructed through the thematic analysis.

3.1 Questionnaire data

3.1.1 Data completeness

Sixty five responses to the online questionnaires were received. In total 11 responses were excluded; seven responses because they were incomplete, and four in accordance with the exclusion criteria, because they reported developmental disorders in addition to CHD (see Figure 2.1). Of the remaining 54 participants only one was missing data for the PWB-PTC questionnaire. The total number of questionnaires analysed was 54.

3.1.2 Participant demographics

Parents were predominantly White British mothers in their mid-thirties (see Table 3.1 for demographics). Parents reported a range of different congenital heart conditions in their children from septal defects to hypoplastic left heart syndrome (see Appendix O for details). Many parents reported co-morbid heart problems.

Table 3.0.1 Demographic features of whole sample parent participants

Variable	Value
Sex	n (%)
Female	53 (98.1)
Male	1 (1.9)
Ethnicity	
White British	51 (94.4)
White Other	1 (1.9)
Mixed	1 (1.9)
Other	1 (1.9)
	\bar{x} , (SD) (range)
Age	36.45 ± 6.71 (25 – 65)

Ninety percent of parent respondents reported their child's last treatment was over one month ago. Nearly one quarter of parents reported their child's last treatment was less than 6 months ago, whilst over half of the parents reported their child's last treatment was over 1 year ago (see Figure 3.1.)

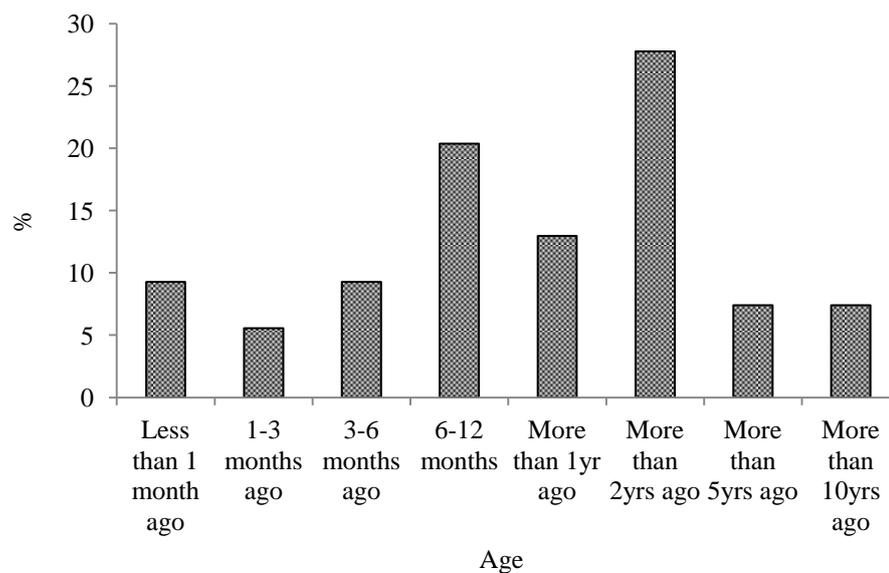


Figure 3.0.1 Distribution of time since last treatment for the whole sample

Current child age and family structure features of the sample are described in Table 3.2. Children's ages ranged from 14 weeks to 17 years. Nearly all children lived with two parents in the house and almost two thirds of households had two or fewer children.

Table 3.0.2 Whole sample child and family features

	n (%)
Child's Current Age	
0 ≥ 1 year	7 (12.96)
>1, ≥ 3 years	9 (16.67)
>3, ≥ 6 years	12 (22.22)
>6, ≥ 10 years	11 (20.37)
>10 years	12 (22.22)
Deceased	2 (3.70)
Missing data	1 (1.85)
Siblings	
None	11 (20.37)
1	22 (40.74)
2	11 (20.37)
3	5 (9.26)
4	2 (3.70)
5	2 (3.70)
6	1 (1.85)
Family Structure	
Two parents	49 (90.74)
Joint custody	1 (1.85)
Single parent	2 (3.70)
Single parent & also sees other parent	2 (3.70)

3.2 Research Question 1: Traumatic stress symptoms

Traumatic stress symptoms were assessed using the PCL-5 questionnaire described in section 2.5.3. The median PCL-5 score of 18.5 (IQR=10.0 – 29.75) for the whole sample of self selecting parents was below the suggested PCL clinical cut off of 33. Twelve participants (22.22%) scored at or above the cut off score. However for diagnosis of PTSD the DSM-5 specifies: one symptom each from criterion B intrusions, and criterion C avoidance, and two symptoms each from criterion D negative alterations in cognition and mood and criterion E alterations in arousal and reactivity (APA, 2013). Using these criteria to make a provisional diagnosis, 13 participants (24.07%) would meet criteria for PTSD.

3.3 Research Question 2: Posttraumatic growth

PTG was assessed using the PWB-PTCQ questionnaire described in section 2.5.4. The average score for the whole sample of 65.57 ($SD=9.56$) was above the cut off score of 54, suggesting on average, positive posttraumatic changes in relation to their child's CHD. Scoring below the cut off suggested negative changes and only five participants (9.0%) were scored below the cut off.

3.4 Coping flexibility

Coping flexibility was measured using the PACT scale described in section 2.5.2. The whole sample median score was 0.83 (IQR= 0.78 – 0.88). This was higher than the average score of 0.79 ($SD=0.11$) found in conjugally bereaved adults with complex grief, but lower than the scores for married ($\bar{x}=0.84$, $SD=0.07$) and asymptomatic conjugally bereaved adults ($\bar{x}=0.84$, $SD=0.08$) (Burton et al., 2012).

3.5 Discussion group data

3.5.1 Participant drop out

Twenty-five participants were entered into 3 discussion groups however one participant dropped out after the second week. This participant's comments in the online discussion were withdrawn from the study at their request (see section 2.9.3). The final sample consisted of 24 participants across 3 groups.

3.5.2.1 Parent demographics and child CHD features

The discussion group sample consisted of 24 White British mothers and one father. Of these, 20 mothers contributed to group discussion. The average age of parent contributors was 35.2 years (SD=5.71) age ranged from 25 to 46 years. Child age ranged from 10 months to 12 years old. Time since last treatment, presented in Table 3.3, ranged from less than one month ago to more than 10 years ago. Nearly all contributing parents reported two parent families. Two parents described blended families and one additional parent reported joint custody arrangements with child spending equal time with each parent. The majority of parents (80%) reported at least one additional child in the family, the number of siblings is reported in Table 3.3.

Table 3.0.3 Time since last treatment and number of siblings for children of parents contributing to group discussion

	n (%)
Last treatment	
Less than 1 month ago	1 (5)
3 - 6 months ago	1 (5)
6 - 12 months ago	5 (25)
1 - 2 years ago	4 (20)
2 -5 years ago	6 (30)
5 -10 years ago	2 (10)
>10 years	1 (5)
Siblings	
0	4 (20)
1	8 (40)
2	5 (25)
3	3 (15)

Contributing parents reported a number of different heart conditions (see Table 3.4). Half the contributing parents 10 (50%) reported post-natal diagnosis and 9 (45%) parents reported antenatal diagnosis. 1 parent was missing this data. The demographic information collected about each contributing participant and their child is presented in Table 3.4.

Table 3.4 Parent and child demographic features for each participant contributing to group discussion

Pseudonym	Parent age	Child current age	Child heart conditions	Diagnosis	Time since child's last treatment	Family structure
Alice	40	3 years	TGA	Antenatal	2 - 5yrs	Child lives at home with both parents and one sibling
Charlotte	35	6 years	VSDs and multi level pulmonary stenosis. Following OHS -free flowing pulmonary regurgitation and pacemaker.	Post-natal	2 - 5yrs	Child lives at home with both parents and one older sibling
Christine	38	4 years	Pulmonary streaks leading to hypoplastic right heart syndrome	Antenatal	2 - 5yrs	Child lives at home with both parents, one older sibling and two younger siblings
Diane	46	8 years	Sub aortic stenosis	Post-natal	1 - 2 yrs	Child lives at home with both parents and 2 siblings
Elizabeth	47	10 years	ASD, VSD, TGA ,CoA	Missing data	2 - 5yrs	Child lives at home with with mum and step dad and two older siblings. Child also spends time with father regularly.
Eva	34	3 years	CoA and aortic valve stenosis	Antenatal	2 - 5yrs	Child lives at home with both parents and one sibling
Helen	28	3 years	Double outlet right ventricle, VSD, ASD , Aortic arch	Post-natal	2 - 5yrs	Child is an only child and lives at home with both parents.
Ivy	40	8 years	AVSD with small left ventricle	Antenatal	1 - 2 yrs	Parents divorced with joint custody. Child is an only child and spends time living with each parent during the week.

Pseudonym	Parent age	Child current age	Child heart conditions	Diagnosis	Time since child's last treatment	Family structure
Jasmine	35	1 year	Tetralogy of Fallot	Antenatal	6-12 months	Child is an only child and lives at home with both parents.
Jessica	31	6 years	CoA, Aortic stenosis	Post-natal	6-12 months	Child lives at home with both parents and one younger sibling
June	30	9 months	VSDs, ASDs, PDA, pulmonary stenosis.	Post-natal	3-6 months	Child lives at home with both parents and two siblings
Laura	35	5 years	Critical aortic stenosis	Post-natal	5 - 10 yrs	Child lives at home with both parents and one older sibling
Leah	25	22 months	TGA, VSD	Post-natal	1 - 2 yrs	Child lives at home with both parents and one older siblings
Lucy	34	4 years	Pulmonary Atresia with IVS	Post-natal	1 - 2 yrs	Child lives at home with both parents and one twin sibling
Marie	34	12	Aortopulmonary window, VSD, ASD	Antenatal	10yrs+	Child lives at home with both parents and one older and one younger sibling
Megan	32	21 months	CoA, VSD, and bicuspid valve	Antenatal	Less than 1 month ago	Lives at home with both parents and 3 older siblings. Child had another older sibling now deceased.
Olivia	30	21 months	TGA, Ebsteins Anomaly	Post-natal	6-12 months	Child lives at home with both parents and two older siblings
Rosie	42	5	Pulmonary atresia and VSD	Antenatal	5 - 10 yrs	Child lives at home with both parents and one sibling

Pseudonym	Parent age	Child current age	Child heart conditions	Diagnosis	Time since child's last treatment	Family structure
Sophie	37	20 months	Plumonary atresia with hypoplastic right heart	Antenatal	6-12 months	Child is an only child and lives at home with both parents.
Vicky	31	11 months	Interrupted aortic arch, VSD, ASD, PDA	Post-natal	6-12 months	Child lives at home with both parents two older siblings and step-sister.

ASD= Atrial Septal Defect, CoA= Coarctation of the Aorta, PDA= Patent Ductus Arteriosus, TGA= Transposition of the Great Ateries, VSD= Ventricular Septal Defect

Table 3.5 Parent and child demographic features for each non- contributing group discussion member

ID	Parent Gender	Parent age	Child current age	Child heart conditions	Family structure	Time since child's last treatment	PCL-5	PWB-PTCQ	PACT
11	Female	30	4 yrs	Tetralogy of Fallot	Child lives with both parents and two siblings	2-5 years	27	72	0.91
33	Male	37	14weeks	Tetralogy of Fallot	Child lives with both parents	Less than 1 month	18	56	0.91
36	Female	38	6 years	Tetralogy of Fallot	Child lives with mother and 5 siblings. Sees father on a monthly basis	2-5 years	11	62	0.78
58	Female	35	2 years	ASD, VSD	Child lives with both parents. Mother is currently pregnant	2-5 years	10	69	0.89

Each contributing participant's questionnaire responses are presented in Table 3.5. The average PCL-5 score for these parents was 21.8 (SD=15.18) and six contributors (30%) scored above the suggested cut off score of 31. Using the DSM-5 criteria to make a provisional diagnosis, four contributors (20%) would meet criteria for PTSD. The average PWB-PTCQ score for contributing parents was 67.9 (SD=10.62). This is above the cut off of 54 suggesting on average, positive posttraumatic changes in relation to their child's CHD. Two participants (10%) scored below the cut off, indicating negative changes. Contributing parents made between one and nine contributions to group discussions which corresponded to 1.2% and 10.5% of the total number of comments in the discussions. The average number of contributions was 4.3.

The demographic information and questionnaire data collected from each non-contributing participant and their child is presented above in Table 3.6. The average age of non-contributing parents was 35 years (SD=3.56). Child age ranged from 14 weeks to 6 years old. The average PCL-5 score was 16.5 (SD=7.85) and no parents scored above the suggested threshold. All parents reported positive changes and the average PWB-PTCQ score was 64.75 (SD=7.18). This suggests the non-contributors may have been less negatively affected by their experiences.

Table 3.6 Questionnaire scores and number of contributions to discussions for each parent

Pseudonym	PCL-5	PWB-PTCQ	PACT	Number of discussion contributions	Group number
Alice	7	79	0.87	7	3
Charlotte	37	77	0.83	4	3
Christine	9	63	0.73	6	1
Diane	42	63	0.76	4	1
Elizabeth	36	84	0.91	2	2
Eva	10	72	0.82	4	3
Helen	29	46	0.72	3	1
Ivy	26	78	0.89	3	2
Jasmine	13	58	0.91	1	2
Jessica	19	77	0.82	4	3
June	41	79	0.8	1	3
Laura	2	67	0.75	6	1
Leah	13	69	0.78	8	1
Lucy	44	51	0.89	6	2
Marie	3	80	0.8	5	3
Megan	51	59	0.65	3	1
Olivia	11	69	0.88	3	3
Rosie	19	71	0.9	5	2
Sophie	14	56	0.83	2	3
Vicky	10	60	0.75	9	1

PCL-5: Posttraumatic stress disorder checklist -5

PWB-PTC: Psychological wellbeing- Posttraumatic Changes questionnaire

PACT: Perceived Ability to Cope with Trauma scale

3.6 Thematic Analysis

3.6.1 Assumptions in the analysis

The thematic analysis had the aims of exploring how parents of children with CHD understand their experiences of raising a child with CHD, in a bottom-up manner. Quotes have been reproduced “verbatim” including all typing errors and shorthand. Pseudonyms and XXXs have been used to preserve anonymity of participants. Quotes are presented in the following format (pseudonym, transcript number: line numbers).

3.6.2 Overview of themes

Three themes and eleven subthemes were constructed through the analysis these are listed in Table 3.7. These are described in detail in turn with quotations from parents’ discussions.

Table 3.7 Summary of themes and sub-themes

Themes	Sub-themes
1. Being a parent of a child with CHD	1.1 The work of parenting 1.2 The “emotional strain it puts on you” 1.3 Extreme distress 1.4 Others not understanding
2. Facing uncertainty	2.1 The “good future” 2.2 Focussing on the now – getting through “one day at a time” 2.3 Sharing experiences
3. “The differences in our lives”	3.1 Family life impact 3.2 Personal changes 3.3 Changes in perspective 3.4 Changes in relationships

3.6.3 Being a parent of a child with CHD

This theme captured the central features in parents' descriptions of their experience of being a parent of a child with CHD. This theme contained four sub-themes which described: the work of parenting a child with CHD, the emotional strain experienced as a parent of a child with CHD, the extreme distress experienced by parents facing the possibility of their child's death, and feeling that others did not understand their experience.

3.6.3.1 The work of parenting a child with CHD

This subtheme described some of the multiple strands of the role of a parent of a child with CHD and the impact of this on parents. Parents' descriptions of how the relational tasks of bonding and their expectations of parenthood were impacted by their child's CHD were also contained in this sub-theme.

Protecting one's child with CHD was described as a strand of the parental role complicated by the child's CHD, for example: "As a mother all I want to do is protect my boys, but he has an added risk factor that I cannot protect him from. This is what upsets me." (Olivia, 3:169-170). In some cases this was described as ensuring protection against poor care and in other cases parents described protecting their child from psychological impingements such as worries, for example: "I'm more of a worrier than ever before taking on the worry of life for my child and not passing it on them" (Diane, 1:399-400). As Olivia states above, when parents perceived they had not been able to fulfil this protector role adequately it was linked to "upset" and, as described by Eva below, "guilt":

I seen [*sic*] him after his coarctation operation still ventilated and unable to control his temperature. I couldn't look at him or stay with him that day, I felt like the worst mum ever! I still have guilt [*sic*] feelings for not being strong for him

that day. I don't think in the future this would happen again..... I'm stronger now!

(Eva, 3:241-243)

“Coarctation” refers to a condition where the aorta is narrowed ranging from mild to severe. “Ventilated” refers to the use of a breathing machine that helps the child to breathe and connects the windpipe via a breathing tube usually through the mouth to the machine. Eva’s description constructs being “strong” through these difficulties as an important quality of a good parent and highlights the implicit motivation to be good parent.

Parents also described a strand of the role of a parent of a child with CHD as facilitating medical care. This included watching over and actively assisting with distressing procedures when necessary, for example:

he had a tube in his chest to drain the fluid build up from the op [*sic*]. When the tube had to come out he protested, so the nurses and I had to hold him down while they took it out. He was writhing and screaming while I held his arms down and the whole thing looked and felt like a torture scene, though of course it wasn't. The next day I went to work but as I was climbing up the stairs to my office my hands started shaking violently and I realised tears were pouring down my face. I ducked into the ladies loo and tried to compose myself, but this was delayed shock from the previous day over which I had no control and I had to wait until my body had finished 'reacting'. (Ivy, 2:80-83).

In addition to describing the active role she played in her son’s chest drain removal Ivy also describes the subsequent impact on her of this experience as “delayed shock”.

Ivy’s description of the uncontrollable physical reaction constructs facilitating medical care as extremely distressing at times.

Another aspect of facilitating medical care was holding responsibility for performing specialised care tasks once the child had returned home for example: “I was taught how to NG tube and sent home to see how she got on” (Marie, 3:129-130) “NG tube” is likely to refer to nasogastric tube feeding (feeding a child directly into the stomach through a tube through the nose). Parents also described monitoring their child at home, “we had to weigh her and take her sats twice a week to watch for any changes” (Sophie, 3:109-110). “sats” is likely to refer to monitoring the level of oxygen saturation in the blood as this can be compromised in children with CHD. Parents also described sharing medical information with others. Facilitating medical care would have required parents to: acquire knowledge of their child’s condition and treatment, learn practical medical skills, and ultimately to take responsibility for their child’s survival by carrying out these necessary procedures.

The tension between occupying the roles of a parent and a healthcare professional was also described by parents. This was interpreted as an additional responsibility on these parents as their professional knowledge intersected with the role of protecting their child, for example:

I also felt like a failure that as a nurse I didn't notice my baby was in severe heart failure. I felt I should have seen how ill he was. I'd spent the previous 2 weeks trying to convince people something was wrong and afterwards I questioned whether I'd tried hard enough (Laura, 1:162-164)

In Laura’s statement the assumption is that her professional knowledge should have informed her ability to protect her child, which compounds her sense of responsibility and associated feelings of guilt. This intersection of roles was also described as extending to protecting partners:

During the bad times I kept some things from my husband - he didn't entirely see the seriousness of some things (as a nurse I saw things differently) and I decided to protect him. I know now that made it worse for me. I felt I was carrying everything myself and had to hold it together for the sake of my family. (Laura, 1:283-286)

Laura's description extends the protector strand of her role of mother of a child with CHD to protecting the whole family and at the same time isolates her in carrying this burden.

Some parents also described efforts to limit the negative impact of their child's heart condition on their child's life. For example "Developmentally he is meeting all expected milestones and *we don't let his heart condition hold him back.*" Lucy (2:18 italics added). In addition to describing a facilitative attitude, Lucy uses meeting developmental milestones as a way to measure the impact of her child's CHD. Through this yardstick she describes her son's development as within normal limits. Similarly Diane describes her efforts to support her son to lead a happy and fulfilled life:

He tries a lot more activities than his siblings did at this age as I want him to try and do everything he can in life and fulfil his life and find an activity he finds he can do well. (Diane, 1:401-403).

This desire to counteract and limit the negative impact of CHD in their child's life was constructed as a strand of the parental role.

Parents also acknowledged the changed experience of the expectations of parenthood with a child without health complications as part of the difficulties experienced in having a child with CHD. For example in a list of distressing experiences Lucy describes: "having twins, and not being able to hold them together, not having twins and getting congratulation cards- we went straight to Mass cards." (Lucy, 2:98-100).

Lucy's expectations of holding her newborn infants together and receiving congratulatory wishes has been replaced by "Mass cards" which was interpreted as cards for prayers for their children. The need for prayers was taken to suggest uncertainty and potential threat to the child. Parents also identified separations and particularly separation immediately after birth as significant for example "After my son was born I held him for 5 minutes or so and then we were separated for 3 days whilst I was quarantined! It was awful!" (Eva, 3:238-239). This was interpreted as the loss of fulfilment of the expectation to hold and bond with one's child immediately after birth. Other separations such as during surgery and other situations where parents were not able to be there for their child were also noted as difficult experiences.

Related to separations parents also described moments of connection and bonding with their child in the development of attachment, for example "I did have times where I felt as though it was just me and him and I would spend hours stroking his thigh (the only bit of him I could touch that did not have tubes coming out)" (Alice, 3:324-326).

However the expectation that the process of bonding would be effortless and unfettered was also not always the met as Olivia described:

Until a year ago I had almost subconsciously not let myself bond with my son for fear of losing him sounds crazy but it's like I was protecting myself from the pain.

Once he had been home for a long period as he grew we have the best bond and the thought of him having to go into hospital again breaks me. (Olivia, 3:313-316)

Olivia describes how the uncertainty of her son's survival complicated bonding with him. How and when parents might expect to hold and bond with their infant, were changed by their child's CHD. *The work of parenting a child with CHD* acknowledged the multiple strands of the parental role for parents of children with CHD and the sometimes

distressing nature of the role as well as the changed experience of the usual expectations of parenthood.

3.6.3.2 *The “emotional strain it puts on you”*

This sub-theme captured parents’ affective responses, feelings, and emotional displays associated with their difficult experiences of their child’s CHD. This sub-theme was deeply connected to the previous sub-theme of the work of parenting as the emotional strains described were often in response to the work of parenting a child with CHD and the multi-stranded role of parents. Parents described affective, physiological and cognitive responses. They labelled feelings such as: anger, guilt, frustration, helplessness and isolation in their experiences and they reported feelings of anxiety centred on their child’s well-being, for example Leah describes:

I think my immediate reaction to the situation we were put in was tears and awful thoughts that my son was going to die. That it was something I did. Guilt. A lot of guilt for it not being recognized and diagnosed before that. Once my son was stable and we were told his diagnosis, apparently I appeared very calm and unfazed. Apparently I seemed to handle it very well and maintained my usual self. Inside I didn't feel this way at all and I know my partner probably knew this. All I felt for weeks was immense pain. Like I was having a permanent panic attack. And I was angry. For me and my son. I assumed that was normal to feel though. I did give me a surge of strength. After all, he was too small to be heard so I had to do it for him! Long term effects, I was told I most likely have PTSD now. I was advised to seek some counselling, if I'm honest, but I haven't. I have off days, mainly of guilt and then anger about it. (Leah, 1:351-361)

Leah, like many parents in the sample described cognitive responses in “awful thoughts” which centred on the fear of losing her child. She highlights the difficult affective

responses she describes as “tears” and “immense pain” which she likens to “a permanent panic attack”. Some of the feelings parents experienced like anger and guilt, as identified by Leah in this example, were constructed as a common and normal response to the acute and intense situations they had experienced. Leah further describes a use for the anger she felt as a “surge of strength” and her description of her son as “too small to be heard” constructs her role to protect her child, in a process by which anger fuelled this drive. Leah’s description contrasts her internal state to her external emotional display which suggests that these affective states and feelings sometimes remained unseen by others and this connects with descriptions from many parents of putting on a “brave face” discussed in the theme of focussing on the now in section 3.6.4.1. Leah’s description of “off days” as a long term effect constructs the experience of these emotional strains as persistent beyond critical events and this was common experience among parents. In addition to parents’ descriptions of feelings of guilt linked to a sense of responsibility to protect and care for their child, feelings of guilt were also linked to sense of responsibility for causing the difficulties their child was experience for example in Leah’s comment “that it was something I did”.

Feelings of helplessness were also described frequently. Sometimes this was in connection with not being able to carry out the parental role “I found the most distressing thing, is the feeling of helplessness. Not being about to do anything myself for my baby.” (Olivia, 3:164- 165). Other times feelings of helplessness were more general and related to feelings of lack of control and uncertainty:

there are a few things that i found quite disturbing/distressing..... the biggest was probably the constant feeling of helplessness and lack of control. so much was happening to us or around us and I had no influence over it and for the most part

didn't really understand (or have the time to process) what was going on. (Marie, 3:180-183)

This highlights the connection between the sub-theme emotional strains and the theme facing uncertainty which is discussed in section 3.6.4.

Parents described persistent feelings of worry and anxiety about their child's health for example:

For the first couple of years she was very sickly and I was quite neurotic and overly anxious about her health and pretty much lived at my GPs surgery.

I am now much calmer overall but still get anxious leading up to check ups.

This is when I slip back into the sleeplessness and worrying about 'what ifs' this tends to only last a day or two now. (Marie, 3:355-359)

In addition to physiological responses and worry Marie describes how her anxiety about her child's health was manifest in relational behaviours by seeking medical opinion and possibly reassurance in many trips to the GP. However this worry was constructed by some parents as justified "Because of the experiences. I find myself worrying overly about my son, but often warranted." (Leah, 1:365). This was described as due to the common experience of susceptibility to respiratory illnesses in winter and the common experience of quick escalation of smaller issues into serious complications for example:

"Every day is tricky and throws new challenges , last winter a simple cold ended up in our daughter Being rushed to hospital as a floppy baby ! And then over Easter couldn't get her to eat , another hospital stay followed my[sic] heart medication." (Helen, 1:204-206).

Thus persistent feelings of anxiety and worry were described as a response to continued real threat for many parents. The experience of worry and anxiety was therefore deeply connected to the strand of protector parent role in the work of parenting.

Parents also described negative feelings of isolation and loneliness during critical times. Parents described being in hospital on their own and separated from family and friends for example:

I would have spoken to more people. more families on the wards. With kids at home hubby was there I was alone with a very poorly 6 day old and although staff were excellent. I regret not approaching another mummy or daddy in the same boat as such for a chat or a cuppa. Lonliness [*sic*] fuelled the fear that we were going to lose XXX too. (Megan, 1:231-235).

Marie also described feelings of isolation as part of the difficulties she experienced in the transition home after surgery, ending with:

I wish at the time I had someone to talk too who had gone through what I had and understood and could normalise things and help me see I wasn't the only one going through this would have helped me feel less isolated. (Marie, 3:223-226)

However isolation from others was also constructed as an active process of limiting or controlling the external environment "I wanted it be just me and my child and shut everyone else out. I was exhausted being such a control freak." (Diane, 1:346-347). This suggests that for some parents being with others could also be experienced as unhelpful or overwhelming at the time.

Parents also labelled negative feelings of frustration in relation to their experience of other parents' and grandparents' child health concerns, for example:

I get frustrated when people tell me how hard it is watching thier [sic] lil [sic] ones have their routine jabs, how bad they feel etc. It annoys me because I wish those were my only concerns, but I try to let it wash over me, as I was once them, with my first, and i wish those were my only concerns now [smile emoticon]
(Leah, 1:216-219)

Leah's response to these emotions suggests reflection and a sense of compassion help her to tolerate the feelings of frustration and annoyance and allow her to let them "wash over" her. Leah's identification of her "wish" to have another's concerns instead of her own was suggestive of a painful and unnamed loss of the expectation of a healthy child. The feelings of frustration parents described were also linked to experiences of others not understanding described in section 3.6.3.4.

One parent described the experience of coping with grief after losing a child late in pregnancy:

Getting through the death of my older son was a whole diff [sic] ball game. Its [sic] a continual ongoing process that i couldnt [sic] describe how i did it...i didnt [sic] cope...i just learn to deal with it everyday a little more since then and find happiness in the laughter and smiles of my other children. (Megan, 1:236-238)

Megan describes the emotional process of grieving as a very different process from the emotional process of coping with the experiences of raising a child with CHD. Overall *the emotional strain it puts on you* was described as a difficult and primary experience deeply connected to the role of being a parent of a child with CHD.

3.6.3.3 *Extreme distress - “Overall the whole experience has been traumatic”*

This sub-theme described parents’ experience of facing the life threatening nature of their child’s heart condition. The extraordinary nature of these experiences was constructed as difficult to put into language and parents described their experiences as incomprehensible, overwhelming, and some parents also reported they had not consciously reflected on all they had been through in the course of their child’s condition for example: “I’m not sure about reactions. Maybe factoring in that most of us have dealt with *unimaginable stress*, and reactions weren't necessarily conscious. I've actually not sat and thought about how I coped/reacted with XXX's experiences until your questions.” (Lucy, 2:142-144, italics added). Lucy’s response highlights unconscious reactions during critical times and give a sense that parents may not have had the space to reflect on their own experience of their child’s condition due to the unimaginable stresses and tasks of the parental role they faced during critical times.

The sub-theme *Extreme distress* was described in the context of parents’ experience of the life threatening nature of their child’s heart condition and included parents’ fears about their child’s survival, for example “The other most distressing aspect of having a chd kid has been my fear of having to guide him through his own death, should he die before me.” (Vicky, 2:88-89). Parents also described facing critical events with a clear immediate threat to life such as: “cardiac arrest”, “crashing”, “stroke”, or a child not breathing, for example “XXX went for a routine cath [*sic*] that was supposed to take an hour, and crashed in the middle of it” (Rosie, 2:101-102). The “cath” is likely to mean a cardiac catheterisation, a short surgical procedure conducted to observe how well the heart is functioning. Descriptions of events where parents faced the possibility of their child’s death were sometimes sparse, stated in matter of fact ways and used medical

language. This could be thought of as use of the defence mechanism of intellectualising to focus on the technical situation rather than the emotions that it brings to mind to manage an emotionally charged topic. Other parents emphasised the extreme difficulty in facing the possibility of their child's death and many of the responses coded in this theme were in response to question two in the schedule asking about their most distressing experiences, for example:

The most distressing times for me was when my daughter was having her second OHS surgery. We had a call from a doctor after only a few hours to tell us to come to the hospital as the surgeon wanted to speak to us. We were about 10 mins walk away and we ran all the way to PICU. I thought they were going to say she had died. I barely felt I could breathe and was like everything was in slow motion.

(Sophie, 3:228-232)

“OHS” refers to open heart surgery, where the child is placed on a heart and lung bypass machine and the heart is stopped during surgery. “PICU” refers to the paediatric intensive care unit. Another parent described “At the time to be told your child might die was *mind blowing* and very *surreal...*” (italics added Vicky, 1:31). Vicky's language illustrates the incomprehensibility of the experience to one's self. Parents also described how incomprehensible this experience was for others for example:

I struggled with the comments of others who simply did not get how serious it was. I was told several times 'oh my child's got asthma and the doctors always make it sound worse, they're fine really' - I felt like it demeaned my experience or that my reaction was over the top. I gave up explaining 'no, he nearly died' (Laura, 1:269-272)

The depth of impact of parents' experience was interpreted from parents' descriptions of the length of time and the intensity of the distress related to their experiences that they continued to suffer. Parents reported long lasting impact from their experiences, for example Elizabeth described the most distressing aspect of her experience as: "My son arresting .. Horrid .. Still stays with me ten years later" (Elizabeth, 2:114-115). Anniversaries were also constructed as difficult, for example:

It will be 1 year to the day on Sunday that XXX collapsed and my whole world changed, and how I thought I had dealt with things coming up to the anniversary of the event is stirring up big emotions. I even broke down in work today as I was thinking this time last year I was holding my new baby and then it he was taken off me for 6 weeks. (Vicky, 1:78-82)

and "The anniversary is always a very reflective time for me, it's been 6 years so no longer upsetting but my family know I find that week difficult" (Laura, 1:92-93). While Laura describes a reduction in the distress with the passing of time she still notes the anniversary as a difficult time.

Related to descriptions of the time course of distress some parents also described a delay between the occurrence of critical incidents and their emotional response to these events for example: "I think you do what you have to, to get through at the time, but can fall apart when it is all over, which may be days, weeks or months later. (Rosie, 2:107-108). Similar to Lucy's comments above about dealing with unimaginable stress, Rosie's description of the necessity of "getting through at the time" overlaps with processes described in the theme "focussing on the now" discussed in section 3.6.4.1. The need to focus on the tasks of ensuring the wellbeing of the child and whole family, may have delayed the processing of the emotional impact on parents and thus might also coincide

with a transition home when parents can be more isolated from sources of support as identified by Marie in section 3.6.3.2. and this might have had an effect of compounding the distress experienced by some parents.

The sub-theme extreme distress was also constructed from the language parents used to describe their response to their experiences. Parents used the terms: “traumatic”, post-trauma and “PTSD” like Leah above and Olivia: “The impact of this has definitely left mental scarring, seeing things you could never imagine and then that being done to your own child is traumatic.” (Olivia, 3:318-319) and “Yep, I too have severe flashbacks due to PTSD” (Lucy, 2:131-132). Many parents like Lucy described what might be classified as symptoms of PTSD such as “flashbacks”, feeling “numb”, “removed” and in “dream” or “bubble” feeling distressed by reminders of critical times when their child was unwell such as location, smell or sounds, and worries and anxiety about their child’s wellbeing, for example:

...I don't sleep properly for days before a hospital check up. Walking into the hospital where his surgery takes place makes me feel physically sick, I have even thought I may pass out.

At the time of any hospital stays I have to stock my bag up with strong pain killers as the stress gives me really bad migraines. I have to eat as much as I can the day before because I am unable to eat anything when my son is suffering.

I can't buy a particular shower gel as it is the one we used in the hospital and if I smell it I get flashbacks from our time at the hospital and how unwell I felt at that time. Sometimes I think that I'm being over dramatic but then I realise how deeply I have been affected. (Jessica, 3:303-311)

Similar to other parents Jessica describes the intensity of the impact of her experiences in physical manifestations of distress commonly recognised as symptoms of PTSD.

3.6.3.4 Others do not understand- “nobody seemed to understand why I wasn’t happy”

This sub-theme described the experience that others did not understand the difficult experiences involved with being a parent of a child with CHD. This was stated directly by parents, for example: “It’s ok talking to family and friends but none of them understand exactly how much of an emotional strain it puts on you. Vicky, 1:76-78). The experience of others not understanding was also constructed from parents’ descriptions of unhelpful comments and advice received from others, for example: “I think some of our extended family don’t quite ‘get it’. Although they are supportive and helpful if needed. We have had the comment “well he is fine now.” Which bugs me!” (Leah, 1:427-429) and “What didn’t help was it’ll be fine” Don’t worry he’s in the best place. Family and friends who just couldn’t understand the actual situation we were in as first time parents (understandably)” (Eva, 3:261-264). Parents attributed the difficulty others faced in understanding their experiences to the extreme nature of their experience described in section 3.7.2.1. This can be captured by Laura’s quote in section 6.4.3.3 describing how other people could not grasp how seriously ill her child was. This sub-theme was connected to feelings of isolation, changed relationships and motivations to seek connection with other parents with similar experiences.

3.6.4 Facing uncertainty

This theme captured parents’ descriptions of uncertainty in their experience of their child’s CHD. Facing uncertainty was constructed as an intrinsic part of parents’ experience of their child’s congenital heart condition due to the variable and unpredictable course of the condition. Parents described experiencing uncertainty about the future and their child’s condition for example “For me the most distressing times that I still remember being told about my ‘bumps’ heart condition and the uncertainty of what

would happen.” (Eva, 3:236-237) and “We have some scary surgery still to come, but no firm decisions on what or when as yet” (Lucy, 2:21-22). In addition this theme also collated the flux of parents’ references to: past surgeries, medical complications, good and poor recovery from surgery, periods of good and ill health, and the need for future surgeries often dependent on their child’s development, for example:

At 8 months she had the second surgery (Glenn) which didn't go well and she had a severe bleed which was life threatening so they had to attempt the surgery again a week later. She recovered well and continues to do quite well although she is becoming increasingly blue and breathless. She is nearly 2 and will need further surgery in the next year or two. (Sophie, 3:110-113)

“Glenn” refers to the Glenn Procedure an open heart surgical procedure which is palliative and is carried out to improve circulation of blood to the lungs. Sophie, like many parents in the sample, described several surgical interventions past and future, surgical complications, recovery and deterioration, which indicate a changing and unpredictable illness course and an associated watchful waiting and monitoring of her child’s health for changes over time. Within this theme the prolonged variability and uncertainty was described as difficult to bear. For example:

we were always told she was fixed . But last year things started to change. She is now on meds x3 a day and now needing more surgery ! I Find it all very hard and that constant feeling of waiting for more bad news !! (Helen, 1:10-12)

and

When she was home and 'better' as I have previously said I struggled to cope. I felt like there was a feeling of doom hanging over me, I kept expecting something to go wrong or for her health to deteriorate. (Marie, 3:348-350).

These descriptions of “feeling of waiting for more bad news” and “feeling of doom” were interpreted as stemming from the uncertain and potentially life threatening nature of the condition.

The theme of facing uncertainty was also constructed from parents’ descriptions of not knowing or understanding in contrast with descriptions of knowledge and knowing. Parents described instances where they felt they did not know what was happening such as during critical events such as diagnosis, surgery or emergencies, for example:

a doc came to the room we had been sent, asked us he[*sic*] questions someone else had also done about why we had been sent there and then he just took our son off to another room really quickly, we had no idea why they had taken our son, then all of a sudden the rooms where[*sic*] busy, he had a box over his head. There was blood everywhere, where they were now I know they were trying to get a cannula in, he was floppy and I just remember the day going on and on like this, full of distressing information we didn't understand, many tears by all. What made it hardest was how sudden it was, how we didn't understand how our baby who they said "probably had reflux" was then given this other diagnosis. (Leah, 1:123–130)

In addition to the high levels of distress reported, Leah’s description highlights several aspects of not knowing. Firstly Leah’s wording: “we had no idea why they had taken our son”, “all of a sudden” and the “box over his head” indicate uncertainty from not knowing what was happening at the time, not knowing what procedures were being conducted and why. Leah points to the sudden nature of the change in diagnosis as the element which caused the most distress. The underlying assumption might be that medical diagnosis should be precise and certain rather than changeable which is in conflict with the reality of uncertainty in medical science. From a perspective of not having medical knowledge

and understanding, medical knowledge and understanding were constructed as having the power to potentially alleviate uncertainty and with it distress. However from the perspectives of parents who were also healthcare professionals this was not the case. Two parents who worked in healthcare described their experience in conversation:

Laura: I feel being a nurse impacted on my experience. I know whilst it was going on I was getting messages from non nurse friends saying "it must be so much better for you being a nurse and understanding everything that is going on". At the same time nurse friends were saying "it must be so much worse as you know fully what is going on" - I definitely identified with the latter! (1:290-294)

Vicky: Yes I know that feeling working in the labs you know all the terminology etc and the worse [*sic*] goes through your head. ... Sometimes a little knowledge is dangerous. My brother and his girlfriend came to hospital when XXX was in AE to support us and since then she said that it really scared and upset her how much I knew and how frank the consultant was being as they couldn't fob us off and had to be totally straight with us. (1:295-301)

These descriptions seemed to suggest mixed feelings towards their medical knowledge. Medical knowledge and understanding was described as something that could be seen by others to be "better" and something that would force healthcare professionals to be "frank" and to take parents seriously. At the same time from their perspective as parents working in health care to know was described as "worse" and "dangerous" and even something to protect others from when taken in conjunction with Laura's comments about deciding to protect her husband from her perspective as a nurse quoted in section 3.6.3.1. Medical knowledge was not constructed as alleviating uncertainty or distress by these

parents but was constructed instead as adding to the difficulties experienced. This may be because this type of knowledge does not eliminate the uncertainty and potentially life threatening nature of the child's condition and might actually bring clearer understanding of potential negative outcomes and clearer understanding of the uncertainty in medical science.

Thus facing uncertainty was constructed from parents descriptions of uncertainty about their child's health condition in the future, descriptions of experiences not knowing, and descriptions of medical knowledge. The sub-themes of: the "good future", focussing on the now and sharing experiences, describe different parental responses to uncertainty and will be discussed in turn.

3.6.4.1 Focussing on the now – "get through one day at a time"

This sub-theme collated parents' descriptions of limiting their focus to the present moment in response to the experience of facing uncertainty. Many parents like Marie and Rosie in the examples below, described focussing on the moment to moment and day to day tasks and issues during critical times:

At the time of diagnosis and throughout rest of pregnancy, birth and through operations I felt scared all the time but other than that I felt quite numb. I guess I didn't have time or energy to process what was happening and so focussed on getting through each hour that it was easier to work on auto pilot. (Marie, 3:344-347)

and

I think the only way we were able to get through the really bad times, was not to look to[sic] far into the future and the obstacles we still had to face. If we took it one day at a time, it was more manageable. (Rosie, 2:118-120)

Both parents describe limiting their focus to the present in order to endure difficult experiences. Marie's use of the phrase "work on auto pilot" suggests an intentional disengagement with the overwhelming experiences similar to Christine's use of the metaphor of an ostrich below:

I'm afraid I'm an ostrich.. I know he's ill nd [sic] I stay with him in hospital thro [sic] any procedures but I almost blank it... *I try to stay cheerful and put on a big brave 'I'm coping' face...* It's only once in a blue moon where it seems to hit me like a sledge hammer...when I think thro[sic] the reality of it all... Those I call my down days.. nd [sic] generally only come once every 2/3 months.... I am a firm believer in not worrying till I'm told there's something to worry about (Christine, 1:318-322 italics added)

Christine describes a process of "almost blanking it" during critical times which was interpreted as avoidance. Despite her beliefs in not worrying unnecessarily, she also describes being struck by forceful "down days" from time to time which break through her avoidance of "the reality of it all". In contrast Lucy describes a less conscious avoidance of reflection on the personal impact of her child's CHD through prioritising the needs of others before her own:

I've actually not sat and thought about how I coped/reacted with XXX's experiences until your questions. I have been more concerned about XXX and his twin sister and my husband. I'm not ordinarily selfless I can assure you. In this instance, I think I coped by focusing on everything except myself. (Lucy, 2:143-146)

Lucy's reflections on this process suggest that this was less an intentional approach and more a fluid consequence of managing the work of parenting and the emotional strain in

the face of uncertainty. “Getting on with it” was another example of this “Like all parents with heart children it is traumatic, but you just get on with it.” (Rosie, 2:48-49).

Focussing on the now was also described as a temporary means to delay processing the emotional strains of raising a child with CHD. For example: “I put all my energy into dealing with everything that was going on when everything got back to normal it hit me what we had been through. (Vicky, 1:85-87). Vicky describes a state similar to Ivy’s description of “delayed shock” after assisting with her son’s chest drain removal described in section 3.6.3.1. Rosie also described a process of getting through but later suffering the impact of one’s experiences in section 3.6.3.3. From these representations focussing on the now was in part a necessity in order to endure the emotional strains and carry out the work of parenting a child with CHD.

Parents also described a range of other ways that they found helped them to manage the emotional strain and face the uncertainty inherent in their child’s condition. This included: being positive, seeking information and putting on a brave face. In the example above Christine described staying “cheerful” and presenting the appearance of coping to others alongside focussing on the now. Similarly other parents described: “I put on a brave face to people and still do if I’m honest. (Vicky, 1:337). Vicky also described:

On the outside though I tried to deal with it factually which helped me process it better, I wanted to know all the medical issues and what I didn’t understand I went and researched (sometimes this was and still is a bad thing). (Vicky, 1:329-331)

Information seeking is often classified as an active coping strategy, whilst not without its limitations as Vicky alludes to, it can also be considered as a strategy to reduce uncertainty. However the opportunity cost of information seeking might be reduced

engagement with the emotional substance of the experience. Similarly instances of minimisation and use of overly medical factual language observed in parents' descriptions of their experiences were interpreted as potentially indicating a distancing of the emotional content in the experience. Some parents also described they unexpectedly found relief from the distraction provided by returning to work:

When my mat leave ended I was petrified of going back... In reality going back was the best thing for me. With a few weeks I felt hugely better with something else to focus on other than my son, what he'd been through and what the future held. (Laura, 1:278-282)

The sub-theme *focussing on the now* described the ways in which parents endured the emotional strains of parenting a child with CHD in order to carry out the work of a parenting a child with CHD in the face of uncertainty.

3.6.4.2 The “good future” - “The fear of the future, I hope he grows old and grey.”

This sub-theme represents a push towards imagining and preparing for a good future in response to uncertainty about the future. Contemplating the future was described by parents as a difficult process in the face of uncertainty. Some parents described mentally preparing for the death of their child or mourning during uncertain and critical times during their child's treatment, for example:

At the time when XXX was being resuscitated, and it's horrible to say out loud, I was preparing myself to tell my family and friends that he had died and thinking horrible thoughts about funerals etc and how I would tell my children (Vicky, 1:327-329)

Leah also describes: “I have awful negative thoughts and assume the worst will eventually happen so rather than get my hopes up on these occasions when he is unwell etc I just immediately think the worst.” (Leah, 1:420-422). These and similar descriptions were taken to suggest that for some parents whilst it may be extremely upsetting to think the worst, it is more difficult at times to hope for a good outcome. That is for some parents hope might be difficult to generate and even painful at certain times.

The sub-theme the “good future” was constructed in contrast to and in opposition of worst case future scenarios and also in opposition of the absence of a future, as described by Ivy below:

I tried to take it one day at a time and not think too much about the future. But my counsellor encouraged me to plan for a good future and stay positive. I used to not buy the next size up of clothing until when it was needed, now I order for the year ahead and have started saving for a future where I hope my son will go to university. (Ivy, 2:135-138)

Similarly Lucy describes the support she received from friends in contemplating a good future:

We planned for happier times in the future. My friends actually all created us a happy days list of all the things I had been prattling on about doing when we all went home together. It was very much NOT a bucket list, but a reminder that happy days were going to resume with both twins. (Lucy, 2:123-126)

Lucy’s emphasis differentiates between a bucket list and her “happy days list”. This was interpreted as highlighting the subtle difference in motivation behind a bucket list which focuses on things to do before dying and a “happy days list” which looks beyond a bucket list and extends forward into a good future.

3.6.4.3 *Sharing Experiences*

This sub-theme described parents' desires to receive support from others with similar experiences and the benefits and barriers to sharing experiences. This sub-theme was constructed from the content of parents' explicit discussion about peer support, the style of interactions in discussion group and parents' views of the group discussions.

In contrast to the experience of others not understanding described in section 3.6.3.4, support from someone who understood the challenges presented by CHD through their own experience was described by some parents as desirable for example: "It would have also been helpful to have other parents who had been through situation were around to offer emotional support, perhaps in a peer support way." (Marie, 3:285-287). This is also similar to Megan's quote in section 3.6.3.2 wishing she had in retrospect talked with other parents on the ward more at the time of her son's hospitalisation in order to reduce feelings of loneliness. Similarly emotional support from a partner going through the same experience was constructed as helpful:

I guess getting through those first few weeks was made so much easier having my partner by my side, I'm not sure how I would have coped on my own, I would have, but having someone to just sit with on those long hospital days was important. *Someone else who understands exactly what it was like.* (Leah, 1:239-242, italics added).

Vicky also describes the difficulty in finding a supportive space to share her experiences:

now no one asks about it and doesn't really want to mention what happened and I feel like shouting out and saying but I need to talk about it in detail of how it was and how I felt to process it all. Same with work colleagues people seem to avoid the subject. My husband isn't a talker and I find that difficult too as I want to re-

live it (for want of a better word) to help me process what happened. (Vicky, 1:302-307)

Vicky describes her need to talk about her experiences in order to process them. Thus parents described several potential benefits from sharing experiences with someone who understood first-hand the experience of having a child with CHD such as reducing loneliness and giving reassurance and processing emotional experiences.

The conversational style of the interactions in one group reinforced the idea that support from others with personal experience was helpful:

Vicky: Hi everybody. I just wanted to say that being in this group is really helping me deal with my emotions surrounding my son and his heart condition. It's ok talking to family and friends but none of them understand exactly how much of an emotional strain it puts on you... People try to understand and say but you have to think of the positives that he is still here with us etc etc and I know this but sometimes it just gets on top of you. . . . I was referred by my health visitor for counselling for post natal depression in April but both her[sic] and the counsellor said it was more like posttraumatic stress. Does/did anyone else feel like this?

Laura: Big hug Vicky x I was diagnosed with posttraumatic stress after XXX. I felt the same that I threw myself into getting on with it, surviving on adrenaline I think and then about a month later it all hit. The anniversary is always a very reflective time for me...I'm glad you're finding this helpful. I know it's easy for people to say but I did find time helped. I also felt before there was an expectation for me to be "happy" or "comfortable" with the situation. I now know that isn't completely true -

my feelings just have to be manageable and that felt easier to do (hope that makes sense) Xxx (1:76-99)

Vicky begins by addressing a comment to the other participants; To which Laura then replies directly to Vicky. Laura then offers support, reassurance and advice by offering a virtual hug, sharing her own experiences and ending with xs. The group dynamic and interactional style appeared different to the other groups and a number of parents explicitly described the supportive experience of discussing their experiences in a group as helpful. However this style of interaction was not present in all groups and thus, as would be expected, not all parents expressed this view:

I'm not sure I've found anything helpful, as Rosie has said, theres [*sic*] not been any discussions. It has, as the title suggested, focused on the most traumatic points in our lives. For me, I find that hard to share. (Lucy, 2:170-172)

Like Lucy some parents identified reflecting on the difficulties they faced challenging:

“It's been quite tough at times admitting to others how hard it was. (Marie, 3:377).

Another parent commented on the group format as a barrier to reflection “I think you would get more exchange of feelings on a one to one, I certainly didn't want to open up too much in a group setting.” (Rosie, 2:163-164). Parents also described past experiences of online forums as a vehicle for peer support, highlighting additional factors which influence whether sharing experiences in online forums might be experienced as helpful:

I'm not currently active in any forums - I found others had different priorities/needs to me and there were at times political and bitchy undertones. I left my last group as it was uncomfortable and made me feel worse than help. I know in the future as my son's condition changes I may need to go back to forums but I also have a core group of heart parent friends who I trust so would probably approach them first for help or support. (Laura, low, 486-490)

Thus it appears that how parents perceive the nature of the interaction in peer support forums is important in whether or not sharing experiences is helpful.

3.6.5 “The differences in our lives”

This theme described the changes parents of children with CHD described as a consequence of their experiences. The four sub-themes depict different areas parents identified as changed by their child’s CHD. This included: family life, personal changes, changes in perspective and changes in relationships. These will be described in turn.

3.6.5.1 Family life impact

This sub-theme captured descriptions of the systemic effects of having a child with CHD. This included parents’ reports of the direct emotional and psychological impact on siblings, grandparents, and partners, for example “Watching his twin sister regress, stop talking, eating and back into nappies, then waving her goodbye as she left with my mum. Seeing my husband sob” (Lucy, 2:96-98) and Vicky described:

It's upset my mum and dad a lot and I think more than they actually let on. I find that no one wants to talk about XXX and what I went through, probably in fear of what I would do... I think it's affected my daughter more than I realised as she still talks about it, and I thought she would just forget with her being 4, but she associates things with the time that it happened and how it made her sad. (Vicky, 1:407-412)

In these examples Lucy and Vicky explicitly highlight the deep emotional impact on the whole family network. Parents also described the practical support they received from extended family such as help with household chores and providing care for other children.

Many parents also described interruptions to daily family life such as staying in hospital for long periods of time, “We stayed in hospital until she was fit enough to go to XXX hospital for her surgery - 5 months in all.” (Rosie, 2:43-44). Hospital stays and treatment was also in some cases far from home and this had implications for the whole family network and the routines of daily life:

the realisation that I would have to be a 3-4 hour journey away from my elder child who would be almost 4 at the time of the surgery. I found it hard to come to terms with the fact that I would have to rely on extended family to do all the things that I usually do. I desperately wanted to be by my baby's bedside but I also wanted to be with my elder son and do all the Mummy things I usually did. (Alice, 3:143–147)

In this example Alice describes the dilemma she felt as her family was separated as distressing and it was stated in the context of a number of experiences she considered difficult. Parents also constructed daily life of a family of living with a healthy child with CHD as organised by the child's CHD for example:

yes, my son was relatively healthy but we were still affected by it on a daily basis - through the funny colours he turned or the fact he just couldn't do much and if he did try he wouldn't be able to catch his breath! (Charlotte, 3:290-292)

In addition to the impact on the family Charlotte's comments also highlight the ongoing worry about the child's health for parents described in the sub-theme emotional strain in section 3.6.3.2. The sub-theme family life impact describes how a child's CHD produces a systemic effect on the whole family network and this highlights the importance of taking a systemic view of the child and the parents' difficulties.

3.6.5.2 *Personal Changes*

Parents described a range of different personal changes within oneself due to their experiences of their child's CHD. This included feeling "stronger", "more confident" and "knowledgeable", for example "As a person, I have become *so much stronger* in any situation and *more confident* in dealing with stressful situations." (Olivia, 3:365-366, italics added) and Leah describes "On the plus side I am much more *knowledgeable* about medication, surgery, procedures, hospitals etc." (Leah, 1:424-425 italics added). Laura also described changes in caring and valuing herself: "It also changed me in that I believe I am important and need looking after too. Taking care of myself is not selfish, it's normal and it's necessary". (Laura, 1:443-444). Some parents also reassessed their capabilities and future prospects in light of their experiences:

I think it has changed me as a person and made me angry at first as to why my child. I have dealt with that now and feel lucky to still have my boy. I always wanted to be a nurse but didn't think I could deal with death but now it's made me think that maybe I should follow what I wanted to do and would love to go into intensive care nursing as I think I can deal with anything now. (Vicky, 1:341-345)

Vicky describes the changes she has experienced in terms of phases beginning with anger to her current confidence in her ability to "deal with anything now". It is interesting that Vicky's attraction to intensive care nursing is in contrast to the experience of other parents who experience anxiety and distress at reminders of their child's treatment in hospital. Vicky's description of her new confidence might be taken at face value, alternatively this might be interpreted as a desire to gain mastery over what has been experienced as traumatic by actively exposing oneself to situations that are similar to the traumatic experience, or Freud's repetition compulsion as elucidated by Bibring (1943). Many parents also described being more worried and anxious about their child's health,

as a consequence of their experiences, previously discussed in section 3.6.3.2. Alice also described the importance of constructing something positive that could be gained from her experience of her child's CHD:

Since my son's recovery I have been a lot more proactive when it comes to children's health and I have attended first aid courses. I do a lot more to raise money for heart charities. I have joined the campaign for all babies to be heart screened at birth. I have discussed the treatment my son received with other parents going through similar things and medical students to try and help. I think this is me trying to turn what was a horrendous experience into something that I can manage and sort of turn into a good thing for me. (Alice, 3:332–338)

Alice describes engaging in a very active process of transforming her experience of her child's CHD into something positive for herself and others which contrasts with the other parents' descriptions of personal change. Nonetheless whether through an active process or changes perceived through the course of events parents reported that their experiences had changed their assessments of who they were within themselves.

3.6.5.3 Changes in perspective

Many parents also described changes in their outlook on life as a consequence of their experiences of their child's CHD, for example:

I would say that both me and my hubby, don't worry about small things the way we used to. As long as the children are happy and healthy that is all that matters. We don't look to [sic] far into the future, or worry about upcoming surgeries, we just try to focus on the here and now. (Rosie, 2:151–158)

Rosie describes a change in perspective; the things they used to worry about are now considered "small things". She describes a change in priorities which focus on health and

happiness. She also links coping with the challenges ahead by trying to live in the current moment. Parents also described other changes such as being grateful, “appreciating life”, recognising the fragility of life and using their child’s health crises to put other problems into perspective. Megan describes:

Us as a family.....we are definitely closer from it all. I think after losing his brother and then the miracle he became by battling the odds to stay with us...gave us all a healthy respect for how short life can be and how quickly things can change.....we’re not spontaneous (who can be with 4 kids and 2 full time jobs lol) but XXX and the events that unfolded during and after his birth and surgeries [sic] has made us live for the moment that little bit more. (Megan, 1:391-396)

Similarly Diane describes:

I realise how precious life is and try and "seize the day" more and make more time for things - even a little thing like movie night or games night at home - just filling our memory boxes with lovely occasions. (Diane, 1:397-399)

The changes in perspective parents describe suggest that for these parents, having a child with CHD and particularly facing the extreme distress of the possibility of their child’s death, have led them to confront existential questions and consciously re-evaluate what matters and what is important to them in life.

3.6.5.4 Changed relationships

Many parents like Megan, in the example above described changes in relationships as a consequence of their experiences. For example Marie describes feeling closer as a family:

I feel quite proud that as a family we came through some really tough challenges closer and stronger and this gives me confidence for the future that we will meet

life's challenges head on, and I think this helps me worry less about the future and more likely to enjoy the here and now (Marie, 3:369–374)

Leah also describes:

The experience has made us as a family so grateful [*sic*] for the little things, and appreciate each other all the more. It was stressful for us as a couple but brought us together even closer I think in the end. (Leah, 1:368-371)

In addition to feelings of being closer as a couple and primary family unit described by many parents Laura also describes feeling closer to her own parents:

I'm closer to my parents who have been brilliant. My mum is one of the few people I can be completely honest with. My mother in law falls into the group of people who simply does not get it, has no clue about what really happened. (Laura 1:439-440).

Laura links understanding and feelings of closeness as she describes feeling closer to her parents and then draws a contrast between her mother and mother in law in terms of their understanding of her child's condition and their experience as parents. Olivia also links feeling closer as a family and feeling distanced from other people who did not understand their child's condition or know how to respond to them:

Our family has become closer and we appreciate life. The people around us however many took a step back maybe through fear of not knowing what to say around us of about the situation. (Olivia, 3:361-366)

From these descriptions it is possible to construct feeling closer and drawing closer as a family, who has been through the same experience together, as a process in opposition to the experience of feeling distanced from other people who did not understand the family's experience described in section 3.6.4.1. Parents also described how encountering others

not understanding their experience affected the way they expressed themselves in relationships:

people would visit and say well he looks so well and better when he didn't really and even now say there is nothing wrong with him now he has had his OHS when there are more ops to follow and general worries everyday but I feel they think we are attention seeking if we express our worries (Diane, 1:314-317)

Diane describes feeling unable to share her worries with others who did not understand the nature of her child's CHD. Similarly Leah describes withdrawal from her relationships:

I don't really talk about how I feel with people anymore, I think people think I should be over it, or I dramatise things, especially those who didn't know me before. So maybe I appear a little more closed or cold than I used too. (Leah, 1:422-423).

Both these parents describe a sense of self-censoring in relationships in which parents no longer felt well supported. It is likely that these experiences would also further reinforce the experience of feeling that others did not understand them and consequently feelings of isolation.

Parents of children with CHD reported changes in several areas as a result of their experiences. In many of the preceding examples parents linked the changes experienced with ways of coping with their experiences. Rosie describes "focussing on the here and now", Alice describes her "proactive" activities, Diane describes "seizing the day" and making time for family activities together and Marie describes how feeling closer and stronger as a family helps her to enjoy the here and now and worry about the future less. Thus changes identified were sometimes ways of coping at the time and equally changes

might also be a product of coping for example feeling more knowledgeable might be in part a product of seeking information to cope, and feeling closer as a family might be in part a product of more time spent together as a family. Thus the distinction between coping strategies and posttraumatic changes was not clear cut.

3.7 Summary

In summary the PCL-5 and PWB-PTCQ scores suggested that on average most parents in the self selecting sample reported traumatic symptoms below clinical levels and most parents identified positive changes as a result of their experiences of their child's heart condition. However some parents reported clinical levels of PTSD symptoms and some parents associated negative changes with their experiences. The three themes constructed through thematic analysis of discussion groups held with a sub-group of parents described how parents understand their experiences in raising a child with CHD in terms of the central features of what it is like being a parent of a child with CHD, the experience of uncertainty and managing this experience, and the differences parents recognised in different aspects of their lives as a result of their experience of their child's CHD.

4. Discussion

4.1 Chapter overview

This chapter briefly summarises and integrates the main qualitative and quantitative findings of interest in the study and discusses how these findings can be understood in the context of previous literature. The strengths and limitations of the methodological design and the clinical and research implications of the study are also discussed. The chapter concludes with a reflective account of the researcher's experience of conducting the study and consideration of how the researcher's position may have contributed to the research.

4.2 Discussion of Main Findings

This study asked three research questions which will be addressed in turn:

- 1) Do parents report traumatic stress symptoms related to their experience of raising a child with CHD?
- 2) Do parents report posttraumatic growth related to their experience of raising a child with CHD?
- 3) How do parents understand their experiences in raising a child with CHD?

In summary the three themes developed through thematic analysis (Braun & Clarke, 2006) of parents' accounts of their experiences described: the central aspects of being a parent of a child with CHD, facing and managing uncertainty, and the changes parents perceived in their lives. In brief, the theme *being a parent of a child with CHD* consisted of four subthemes that each captured a central aspect of the experience. *The work of parenting a child with CHD* described the challenges presented by a multi-

stranded parental role and the impact of their child's condition on expectations of parenthood. "*The emotional strain it puts on you*" captured parents' feelings, emotional displays and affective responses to their experiences. *Extreme distress* described parents' responses in the extraordinary context of facing the life-threatening nature of their child's condition. Finally *others do not understand* described parents' experience of feeling that others did not understand their experiences. Parents described *facing uncertainty* as a central feature of the difficulties they experienced and as an intrinsic part of their child's CHD condition. In response to the uncertainty they faced parents were described engaging in processes of: *focussing on the now*, looking towards *the "good future"*, and *sharing experiences* with others. Finally the theme "*the differences in our lives*" described the changes parents perceived in the following four areas: *family life impact* which described the wide reaching systemic impact of the child's CHD on the whole family, *personal changes* which described the changes parents perceived in themselves, *changes in perspective* which described the changes parents perceived in their values and outlook on life and *changed relationships* which described the changes parents perceived in their relationships with others.

The themes described in this study centred on how parents understand their lived experiences and differed from the theory of "Parenting under pressure" (Rempel et al., 2013) which was constructed from grounded analysis of interviews with parents and grandparents of children with hypoplastic left heart syndrome (HLHS). The study by Rempel et al. (2013) conceptualised "parenting under pressure" as involving a sequence of four overlapping and iterative phases across a child's illness trajectory from diagnosis through surgeries to transitions home and subsequent developmental transitions. These phases were named: "realising and adjusting to the inconceivable", "growing increasingly

attached”, “watching for and accommodating to the unexpected” and “encountering new challenges”. Unlike the Rempel et al, (2013) study the themes developed in this study were based on thematic analysis of the experiences parents of children with a range of different heart conditions and focussed on how parents understand their lived experiences rather than seeking to explain the processes of parenting a child with CHD.

4.2.1 Traumatic stress symptoms

This study used the PTSD Checklist for DSM-5 (PCL-5) to assess traumatic stress symptoms in a self-selecting sample of parents of children with CHD. The PCL-5 is a newer measure of PTSD based on DSM-5 diagnostic criteria. In this study sample, the median PCL-5 score was 18.5. This is higher than the average score of 15.42 (SD = 14.72) found in a sample of undergraduate psychology students self identifying as having experienced a “very stressful life event” (Blevins et al., 2015). Additionally 22.22% of the sample in this study scored above the recommended clinical cut off of 33, and 24.07% met DSM-5 criteria used to indicate the presence of PTSD. This is higher than the prevalence rate of 16.0% found using the PCL-5 in an undergraduate population in the Blevins et al. (2015) study with a lower cut score of 31 as part of signal detection analysis. One important difference between these samples might be the type of traumatic event experienced as the “magnitude” of trauma has been identified as a risk factor for PTSD (APA, 2013, p278). Participants in the Blevins et al (2015) study reported a range of traumatic events; the most prevalent were: motor vehicle accident, sexual/physical assault, sudden violent or accidental death of a loved one, life threatening illness and fire or natural disaster which accounted for 86.5% of participants. As the current study did not select parents based on a single event such as diagnosis or surgery, it is difficult to say whether the average experience of the events in the Blevins sample is comparable to the

cumulative, ongoing and wide ranging difficulties experienced by the parents in raising a child with CHD in this study.

The prevalence rates of PTSD in the studies previously described (section 1.5.3) ranged from 3% (Toren & Horesh, 2007) to 16% (Helfricht et al. 2008) using the Posttraumatic stress Diagnostic Scale (PDS). The PDS has demonstrated good convergent validity with the PCL-5 (Blevins et al., 2015). The high proportion of self selecting parents scoring above clinical cut off and meeting DSM-5 criteria in this study may be related to the self-selecting nature of the sample who were recruited through a CHD charity (see section 4.3.1.1 for discussion of the sample limitations). Whilst the current study sample is not representative of all parents of children with CHD, it is still of note that some parents of children with CHD reported traumatic stress symptoms and some parents reported symptoms to a degree that might satisfy current DSM-5 diagnostic criteria.

Parental descriptions of what might be classified as symptoms of traumatic stress and the use of trauma terminology were captured in the sub-theme *extreme distress*. This sub-theme described parents' acute and intense responses to the extreme situations and difficulties they faced in the context of the life threatening nature of their child's heart condition. *Extreme distress* was constructed as a part of *being a parent of a child with CHD* and also included parents' descriptions of the incomprehensibility of this experience both from the perspective of parents and also from the perspective others. Parents were asked directly about their most difficult experiences related to their child's CHD and whilst the terms "trauma" and "PTSD" were not explicitly used in the discussion group

questions, participants had completed questionnaire which referenced traumatic events, which might suggest some top-down contribution to the sub-theme.

The sub-theme *extreme distress* was similar to the theme “facing the possibility of my baby dying” constructed from mothers journal entries around the time of their child’s cardiac surgery (Harvey et al., 2013). However *extreme distress* described a response enduring beyond the time of cardiac surgeries, recurring with reminders of difficult experiences, anniversaries, and changes in health condition of the child. In her exploration on maternal ambivalence Parker (2005) used Klein’s object relations theory of infantile development and conceptualisation of the good internal object to understand how separations and the threat of loss of a child also threaten a mother’s deep sense of internal goodness, giving potency to the experience of separation and threats to the loss of the child for mothers. Thus according to this theory for mothers of children with CHD, facing the life threatening nature of their child’s condition might additionally trigger feelings of threat to a mother’s sense of herself, perhaps explaining some of the extreme and indescribable experiences parents expressed in this study. Parker (2005) qualifies that mothers will vary in their vulnerability to the experience of destruction of their good internal object during times of separation and threat of loss of the child.

Overall parents’ reports of traumatic stress symptoms in this study corroborate previous research findings that construct the experience of having a child with CHD in terms of anxiety and trauma. “Acute stress in the most difficult experience” was constructed from thematic content analysis of interviews with mothers of infants who had recent cardiac surgery (Re et al., 2013). However, this theme is not specific to parents of children with CHD and similar themes have been described by parents of children in

intensive care (Colville & Cream, 2009), parents of children with life-limiting and life-threatening illnesses (Bally et al, 2018), and parents of children with chronic health conditions (Kepreotes, et al, 2010).

4.2.2 Posttraumatic growth

This study used the psychological well-being –Posttraumatic changes questionnaire (PWB-PTCQ) to assess PTG in a self selecting sample of parents of children with CHD. The PWB-PTCQ is a relatively new measure of PTG and its use with parents of children with health conditions has not been reported. Positive changes related to their experience of their child’s CHD were reported by 91.0% of parents in this study with 9.0% percent of parents reporting negative changes. Estimates of PTG using the PTG Inventory in parents of children with other health conditions range from 54.3% of parents of children undergoing surgery for congenital diseases (Li et al., 2012) to 88% of parents followed up 4 months after admission to a paediatric intensive care unit (Colville & Cream, 2009). The average PWB-PTC score of 65.57 found in this study was much higher than the reported baseline scores of 50.82 in a small sample of adults having experienced a range of traumatic events recruited online to an internet based expressive writing study using the same PWB-PTCQ measure (Stockton, Joseph, & Hunt, 2014). One explanation for this higher rating might be informed by critical social constructivist views which point to the increasing emotional value of children and concomitant increasing emotional investment in children in western society (Ambert, 1994). Thus given this emotional investment parents in this sample might be more motivated to make meaning of the events and this might be an importance process in helping some parents to cope. In their phenomenological study on parents caring for a child with a progressive and chronic illness (Gravelle, 1997) described parental love and commitment to their child as an influence on the meaning constructed from facing challenges and adversity. If

emotional investment or meaning and value of the child is seen as a motivating factor in making new positive meaning from difficult experiences this has implications for understanding who is more likely to experience PTG and why. Future research to explore this might use the PWB-PTCQ in different representative populations such as: parents of children with health conditions compared with spouses of individuals with a comparable physical health conditions and the individual with the health condition.

In the discussion groups parents were asked directly about any changes they had experienced as a result of their experiences of their child's CHD. Positive changes were among the changes described in the three sub-themes: *personal changes*, *changes in perspective*, and *changed relationships*, which mirrors tripartite conceptualisations of PTG in the literature (Calhoun & Tedeschi, 2001; Joseph et al, 2005). Reports of positive changes in this study are similar to previous research findings on PTG in parents of children with CHD. Having a child with CHD has been referred to as a "blessing" by parents of children who had cardiac surgery (Wei et al., 2016). The theme "finding meaning and spiritual connection" was constructed in a phenomenological study of journal entries made by mothers of children having complex cardiac surgery and describes mothers finding benefits and changes in perspectives (Harvey et al., 2013 described in section 1.3.2). Similar to the study by (Wei et al., 2016) the current study also found that parents reported strengthened relationships as a result of their child's CHD. Furthermore "a new reality" was constructed from studies of parents with children with chronic health conditions to signify a changed way of viewing the world and living (Kepreotes, 2010). Bally et al., (2018) also constructed the themes: "changing priorities" and "salvaging family relationships" in which parents constructed positive changes in outlook and family relationships as a consequence of the child's life-threatening and life-

limiting illness. Although parents in this study described some positive changes they were not disproportionately positive in their responses and they also described negative changes as a consequence of their experiences such as increased anxiety and worry about their child's health. Additionally while parents reported feeling closer as a family they also reported feeling distanced in other relationships and this was linked to feelings that others did not understand their experience, which presents a more complicated picture of the changes parents perceived in their lives.

4.2.3 Being a parent of a child with CHD

Parents across all societies must protect and nurture their children (Bornstein, 2006). However, parents in this sample constructed a multi-stranded parental role and their expectations of parenthood, as changed and challenged by their child's CHD. That parents described facilitating medical care aspects of the parental role is not unusual. Advances in technology have supported trends toward paediatric home care for children with serious health conditions and while there are benefits for many of the involved parties, paediatric home care has also placed increased burdens and responsibilities on parents (Wang & Barnard, 2004). Parents in this study described the impact of witnessing and assisting with difficult medical procedures and holding the responsibility for ensuring their child's survival as extremely distressing. A changed parental role has also been described elsewhere in the literature on parents of children with CHD (Harvey et al., 2013; Rempel & Harrison, 2007). Aspects of the parental role constructed in this study are similar to the theme of "safe guarding their child's precarious survival" developed from interviews with parents of children with HLHS (Rempel & Harrison, 2007). Using a grounded theory analysis this theme was conceptualised as an overarching aim with similarities in the subthemes of: taking charge, in which parents described taking

responsibility for specialized care tasks; and involving others; in which parents described involvement of the wider family network (Rempel & Harrison, 2007). Similarly constructing a changed parental role the study by Harvey et al. (2013) reported a core theme of “mothering through it all” Both these studies used parents’ voices to construct an idea of what constitutes good parenting in difficult circumstances.

Underlying the expectations of parenthood and constructions of what parenting in exceptional circumstances should look like are wider socio-cultural beliefs, and assumptions about what parenting, and family and childhood are. This study took a critical realist stance which acknowledges that understandings of phenomena are limited by the discourses available to describe these phenomena. Whilst it is beyond the scope of this discussion point to critique these discourses it remains important to recognise critical social constructionist viewpoints which acknowledge the influence of dominant sociocultural norms and the construction of ideas about parenting and the encoding of these ideas in structures in society which regulate parenting (Ambert, 1994). Some of these identifiable influences include a research tradition embedded in a western patriarchal heteronormative society which centres on the nuclear family and the central role of mother, typified by theories such as attachment theory (Bowlby, 1969) which reinforce heteronormative patriarchal family structures in society and reinforce the power of medical and legal social structures to regulate parenting. In this study being a parent of a child with CHD will be influenced by these prevailing cultural norms and discourses as well as discourses on health, illness and childhood. For example children are commonly viewed in popular culture as the hope for the future and children have been seen as an extension of the self or a projection of the better parts of the self which can compound the distress experienced when a child’s survival is threatened. Parkes & Prigerson (2010)

point out that it is only recently that infant and child mortality has become a rare event and that prior to this century the loss of a child was a much common experience, shaping the expectancy of loss. They also make the case that decreases in birth rates in western European societies may also contribute to the rise to prominence of the experience of grief and distress at the loss of a child (Park & Prigerson, 2010).

The emotional and affective responses parents described as part of their experiences of their child's CHD often in response to *the work of parenting* were similar to reports of the emotional turmoil and "strife" described in parents of children with chronic illnesses (Coffey, 2006; Kepreotes). However feelings of anxiety and worry about their child's well being were described by parents in this study as a response to a real and continued threat to the child. Parents in this study also described a loss of fulfilment of expectations as part of their experiences. Similar parental experiences of grief, chronic sorrow, and loss of the idealised healthy child have been reported elsewhere in the literature on parents of children with serious health conditions (Bally, et al, 2018; Kepreotes et al., 2010; Maltby, Kristjanson, & Coleman, 2003; Smith, et al., 2013). Theories of grief have also been applied to the experiences of parents with a child with a serious health conditions (George, Vickers, Wilkes & Barton, 2007; Lowes & Lyne, 2003).

4.2.4 Facing uncertainty

The theme *facing uncertainty* in this study described the intrinsic and ongoing experience of uncertainty for parents due to the variable and unpredictable illness trajectory and the potentially life threatening nature of the condition. This theme corroborates descriptions in the literature of difficulties in the experiences of parents of

children hospitalised for cardiac surgery being the “medical course” or the “uncertain and unfolding nature of the diagnoses and surgery” (Kosta et al. 2015, p1059). Quantitative findings also support descriptions of uncertainty as a stressor for parents. An internet survey of parents of children with CHD (Hilton-Kamm, Sklansky, & Chang, 2014) found uncertainty was rated highly as a stress factor. Rempel et al. (2013) describe uncertainty as keeping parents of children with HLHS in a state of “alertness” and anticipation. Uncertainty has also been described by some as “dominating” the experiences of parents with children with chronic, acute, and critical illnesses, with consequences for parents in: psychological distress, challenges to parental roles, and personal growth (Stewart & Mishel, 2000).

The theme *facing uncertainty* in this study also described experiences of not knowing and medical knowledge. Descriptions of not knowing what was happening during critical and distressing situations have been described in previous research using qualitative interviews with parents of children with CHD in Sweden. Carlsson et al., (2015) reported a subtheme of “difficulties sorting out information when in emotional chaos”. They concluded that the emotional turmoil at the time of diagnosis in combination with acute stress reactions made it difficult for parents to understand information given. However in this study knowledge and understanding from working in healthcare was described with mixed feelings as something both powerful and dangerous which compounded the difficulties experienced. This calls for further consideration more generally of how knowledge and understanding impact on the experience of uncertainty and specifically on how the experiences of parents with pre-existing medical knowledge and experience might be different from those without.

In this study the sub-themes of *the good future*, *focussing on the now* and *sharing experiences* described different parental responses to *facing uncertainty*. Many of the parental responses described in this study have been reported elsewhere in the literature on how parents cope when their child has a serious illness. For example similar to the theme *the good future* Bally et al (2018) constructed “the emergence of hope” as an essential life-sustaining response to the uncertainty associated with having a child with a life-threatening or life-limiting illness. Similar to accounts from parents in this study Kovacs, et al (1985) also reported a quarter of mothers of children with diabetes in their study used mental rehearsal and preparation for their child’s death as a means of coping with distressing news. An alternative explanation of this process from a psychodynamic perspective involves understanding the additional threat to a mother’s own internal good object triggered by the potential loss of the child. Parker (2005) posits that the anxiety and fear loss of child that separation can trigger gives rise to drives to restore internal image of the self as a good mother. In a simplified example of this Parker (2005) described one interpretation of the ritual one mother described in parting from her children in the morning and then imagining their death, as a way of restoring her internal sense of herself as a caring mother in the context of separation.

In this study the theme *focussing on the now* described parents limiting their focus to the present moment in response to the uncertainty they faced. Assuming a finite human attentional capacity (Kahneman, 1973), by attempting to limit one’s focus to the current moment and the current issues at hand parents may have temporarily avoided or suspended the processing of difficult experiences. Stewart & Mishel (2000) constructed two competing but equally adaptive “information management styles” related to managing uncertainty from studies of parents with children with chronic illness more

generally. These were: seeking information through questioning and vigilance, and limiting or discounting information which draws attention to uncertainty. In some ways *focussing on the now* appears similar to the latter strategy although *focussing on the now* tended towards avoidance of emotional processing rather than limiting information about uncertainty. Additionally some parents in the current study described this process as less of an intentional strategy. It is clear that being a parent of a child with CHD can evoke powerful and overwhelming emotions. The defensive strategies of avoidance and denial alluded to in *focussing on the now* and other ways of responding to uncertainty such as putting on an “I’m coping face” may have allowed parents to manage the impact of their overwhelming experiences in order to carry out the necessary functions of the parental role. Thus for parents these responses to uncertainty were contextually appropriate and adaptive. The costs of wearing an “I’m coping” mask might be if it is not recognised as mask and a tool, and as such gives a false impression that may hide the real needs of parents. A false impression may also perpetuate unrealistic expectations from healthcare professionals, and also from other parents of themselves, if parents are not allowed the time and safe space to remove these masks.

The desire for peer support constructed as part of the subtheme *sharing experiences* was similar to the views of other parents of children with CHD in the literature. A US study found nearly a third of respondents in an internet survey of parents of children with CHD reported they would have liked more information on support groups or survivors with CHD (Hilton-Kamm et al., 2014). Qualitative interviews with parents of children with CHD in Sweden reported peer support was wanted (Carlsson et al., 2015). Parents also described reading online blogs describing the experiences of other parents with a child with CHD as helpful (Bratt, Jarvholm, Ekman-Joelsson, Mattson, & Mellander, 2015). The wider literature on parents of children with serious health

conditions also reflects the value of peer support to parents (Bally, et al, 2018; Coffey, 2006; Kepreotes et al, 2010). Stewart & Mishel (2000) have theorised, according to Mishel's uncertainty model of illness (1988), that sharing experiences with other families influences uncertainty as parents are able to form and test tentative models of what they can expect in the future. Participants in this study described no longer feeling alone in their experiences as a positive benefit of peer support so it could be argued that the models of what might be expected in the future proposed by Stewart & Mishel might refer not only to what parents might expect for their child's future but also to what parents' might expect of their own personal experience of their child's illness. In her exploration on maternal ambivalence from a psychodynamic perspective Parker (2005) uses the concept of "mothers as containers for other mothers" p3 and "mothers mirroring mothers" p4 to understand the phenomena of mothers seeking connection with other mothers and finding comfort in the reassurance and company and reflections of other mothers and this might explain both the desire for peer support reported here and the experience of support experienced by some parents in this study. Parker (2005) goes on to note that the mirror of the other mother can contain distortions from personal projections and is also influenced by dominant socio-cultural expectations, and as such rather than containing and reassuring, can be experienced as stressful, and anxiety provoking. This might account in part for the descriptions from parents of difficulties opening up in group settings and difficult past experiences in group forums found in this study. These findings can also be considered alongside findings from the Re et al., (2013) study in which a child psychotherapist conducted qualitative research interviews with mothers of young infants with CHD. They reported 15 out of 26 mothers reported difficulty thinking and talking about their experiences. They also found that mothers reported participating in the research interviews helpful. Re et al., (2013) proposed the research interviews had

allowed space for: reflection, connection, “scaffolding” and “building meanings beyond the experience of trauma” p 283. Re et al. (2013) used the psychodynamic concept of emotional containment (Grotstein, 1981) to interpret the function of the research interview, and this may point to an element missing from the peer support and discussion group formats for some parents.

4.2.5 “The differences in our lives”

The sub-theme *family life impact* in this study corroborates the literature on the impacts on family life on: siblings and family life from the perspectives of parents of children with CHD (Kosta et al., 2015). Grandparents of children with CHD have been described as stepping in as needed and playing a role in safeguarding relationships amongst a triad the affected child, the child’s siblings and the child’s parents (Ravindran & Rempel, 2011). Similarly family impact has been identified as a consistent theme from the perspective of parents of children with other serious health conditions (Bally, et al, 2018; Coffey, 2006; Smith, et al., 2013). That a child’s illness affects the whole family is not controversial and it is likely that there are commonalities in the ways in which families are affected across health conditions for example by having to spend time in hospital away from home. However it is also likely that specific individual impacts based on the ways in which the health condition uniquely affects that child within the unique context of that child’s family. This offers support for the systemic approach of models such as the PMTS and reiterates the importance of a systemic approach in formulating the difficulties for parents, children and their families and the type of support and interventions that might be helpful for the whole family unit.

4.2.6 Summary of implications for application of the concepts of PTSD and PTG in parents of children with CHD

A small proportion of the self-selecting parents in this study met criteria for a diagnosis of PTSD and some parents in the study identified symptoms of PTSD such as “flashbacks” and used terms such as “PTSD” and “trauma” to describe their responses to their experiences of their child’s CHD. The sub-theme *extreme distress* described these parental experiences in the context of facing the life threatening nature of their child’s heart condition. Coupled with the unpredictable and variable illness trajectory of CHD described in the theme *facing uncertainty* the findings from this study suggest that it is not appropriate to apply linear models of PTSD or PTG with a single triggering traumatic event to understand the experiences of parents of children with CHD. For example some models of PTG in parents of paediatric patients have used the simile of the aftershocks of an earthquake to understand the impact of events such as treatment, surgery and complications which cause parents further distress (Picoraro, Womer, Kazak, & Feudtner, 2014). These “posttraumatic after events” can then considered as a continuation of the original trauma or as a new traumatic events. However this implies that diagnosis is taken as the major traumatic event. The findings of this and other studies suggest that parents find a number of situations extremely distressing not just diagnosis. In fact the period of diagnostic uncertainty before a diagnosis is given has been identified by parents as extremely distressing across a number of studies (Stewart & Mishel, 2000). In addition the lengthy and variable nature of the illness trajectory described by parents in this study does not support a linear conceptualisation. Thus findings based on the assumption that diagnosis is the original traumatic event might underestimate the contribution of subsequent events to parents’ experience of distress and also the effect of cumulative experience of events on parents’ level of distress. Additionally taking diagnosis as the

original traumatic event for parents of children with CHD potentially confounds severity of condition as more severe forms of CHD tend to be diagnosed earlier eg antenatally. Thus the findings from this study support the individual consideration and formulation of the unique characteristics of the medical illness, and individual contexts, which influence the experience of traumatic stress symptoms as described in the model of PMTS {Kazak et al, 2006}. Thus the findings of this study also lend support to both the PMTS model (Kazak et al., 2006) and the ADD model (Papadopolous, 2007) of posttraumatic reactions which both propose that traumatic events and responses to trauma might not follow a linear pattern. The PMTS model proposes that potentially traumatic events might be recurrent or cyclical (Kazak et al., 2006) whilst the ADD model (Papadopolous, 2007) describes the ongoing difficulties as “facing adversity”. This might fit better with parents’ experiences due to the uncertain nature of their child’s illness trajectory and treatment outcomes. A fuller description of parents experiences might be best captured by elements of both models: parents seemed to be facing ongoing adversity and traumatic events.

Many parents in the sample reported some positive changes as a result of their experiences however, parents in the discussion groups often referred to changes they experienced and ways of managing their experiences within the same statement and it was at times difficult to determine whether a change in behaviour aimed at managing the experience would be classified as a coping strategy or a change experienced as a result of the traumatic experience. This finding reflects current debates in the literature on whether PTG is a coping strategy or an outcome following a traumatic event (Park & Helgeson, 2006). Additionally as described in section 4.2.2 the mixture of changes that parents of children with CHD described in their lives as a consequence of their experiences extend beyond the changes captured by the construct of PTG.

4.2.7 Summary of implications for a non-categorical approach in understanding the experiences of parents of children with CHD

Many of the findings from parents of children with CHD in this study are similar to the experiences of parents across health conditions. For example commonalities can be identified in: the overwhelming emotional experience, a changed parental role, the central and difficult experience of uncertainty, the wider systems impact, and the positive changes parents perceived resulting from their experiences. Thus the findings of this study lend support to a non-categorical approach to understanding the experience of parents of children with health conditions more widely.

The results from this study suggest that specifically for parents of children with CHD it is important to recognise that the experience of uncertainty for parents is bound up with the nature of the condition. To begin with CHD encompasses a diverse range of conditions from mild to severe and life threatening. The illness trajectory can be unpredictable, treatment plans can change dependent on the child's development and in some cases surgery is palliative rather than corrective. Treatment can involve a series of surgical procedures beginning immediately after birth. Thus how uncertainty presents in each individual heart condition, and each individual context, would be best mapped out in a shared individual formulation.

4.3 Methodological strengths and weaknesses

Like all research, the design choices made in this study confer strengths and limitations on the work which must be considered when interpreting the findings and

considering the implications of the research. This section will provide a methodological critique of the work.

4.3.1 Study design

The current study took a critical realist epistemological stance and used a priority sequence mixed methods design with the aim of using multiple descriptors to situate and contextualise parents' accounts of the impact of the difficult experiences in raising a child with CHD. Of particular interest were the constructs of traumatic stress symptoms, and PTG. From within this epistemological stance, one of the strengths of this study can be considered its methodological pluralism in exploring these constructs. Such forms of triangulation have been critiqued as originating from a realist position which assumes some fixed reality which can be referenced (Barbour, 2001). However this is not incompatible with a critical realist position, which maintains a realist ontology. Additionally triangulation can be thought to offer comprehensiveness and a means of reflexivity in analysis of the data (Mays & Pope, 2000). This is not without its challenges and requires a degree of tolerance of uncertainty whilst holding in mind the aggregated findings from questionnaires and qualitative accounts from groups of parents with different levels of coping flexibility which can be difficult to integrate.

4.3.1.1 Sample

4.3.1.1 Selection bias

There were a number of aspects of the study that indicate a potential for selection bias. Firstly the self-selecting participants were recruited from a charity which provides information and support to parents of children with CHD. Thus it cannot be assumed that the sample was representative of the general population. Secondly, the participants must

have sought out the organisation in the past, presumably for support again limiting the generalisability of the findings. Thirdly, the online format of the study excluded parents of children with CHD who do not have access to the internet or who do not use it to access support. However past research suggests that parents of children with CHD do use the internet to access information about their child's condition (Ikemba et al., 2002). Finally, the current study also excluded parents of children with developmental conditions, as it was thought that in addition to the CHD these parents may be managing and coming to terms with their child's developmental difficulties. However children with CHD may be at risk of developmental delays (Mussatto et al., 2014). Taken together this might suggest that the experiences of parents not included in the study may be different from the study participants, which might bias the findings.

Another limitation lies in the representativeness of the sample. The sample was predominantly White British mothers. In a meta-analysis ethnicity was found to moderate the relationship between PTG and measures of global distress so that high PTG was associated with less distress when the study population was made up of 25% or more of people from ethnic minorities (Helgeson et al, 2006.). PTSD (APA, 2013) and PTG (Vishnevsky, Cann, Calhoun, Tedeschi, and Demakis, 2010) also have a higher prevalence rates in women than in men.

Previous qualitative research on the experiences of fathers of children with CHD also suggests that fathers' experiences are different from the experiences of mothers (Clark & Miles, 1999; Wei et al. 2016). The experiences of fathers were not captured in the qualitative element of this study. Overall the conclusions of the study might be better stated as related to the experience of mothers rather than "parents". Future research would

need to overcome fathers' barriers to participation or limit the study to mothers; both these approaches have their individual challenges.

Participants were also a varied group. The recruitment criteria did not specify the: age of child, heart condition, severity of the heart condition, number of or type of surgical procedures experienced or time since last procedure. This would apply to the estimates of traumatic stress symptoms and PTG. However given the research aim to investigate the experience of raising a child with CHD in its entirety rather than a specific aspect of the experience, variation in the population was allowed for. Qualitative research often emphasises ensuring that diversity of variables and contexts thought to influence the phenomena of interest, are reflected in the sample (Mays & Pope, 1995). Thus variability in the sample is not necessarily a threat to the soundness of the qualitative research.==

4.3.1.2 Quantitative measures

It was hoped that the quantitative element of this study could contribute to the research literature to support the investigation of traumatic stress symptoms and PTG in parents of children with CHD with newer measures such as the PCL-5 and the PWB-PTCQ. However the sample in this study cannot claim to be representative of the wider population of parents of children with CHD. Additionally one weakness of using newer measures is the lack of appropriate norms with which to contextualise the findings.

This study used self-report measures to assess traumatic stress symptoms in the study sample. Self-report measures have been found to overestimate the prevalence of PTSD in mothers of children with cancer in comparison to estimates based on gold standard structured clinical interviews (Stoppelbein & Greening, 2007).

This study used the PACT to assess coping flexibility in parents to facilitate purposeful sampling of parents with different ways of coping. The PACT measure has not been used widely in the literature and while it references coping with traumatic events the narrow trauma focussed and forward focussed coping scales did not seem to capture the wide range of coping strategies that parents described in the qualitative discussion groups. Thus it may not be the most appropriate measurement tool for this sample. Furthermore the future oriented nature of the measure makes it hard to understand what perceived ability to cope with trauma actually measures and how this relates to actual coping with potentially traumatic events. Future studies could weigh the benefits and costs of using an additional measure of coping alongside the PACT to allow further assessment of what the PACT actually captures and its utility and relevance in this population.

4.3.1.3 Qualitative research methods

Online focus groups were used in the study to gain an understanding of the experiences of parents raising a child with CHD in a UK sample and specifically related to the constructs of: traumatic stress symptoms, and PTG. The strengths and weaknesses of this part of the study will be discussed with reference to the four criteria for assessing the soundness of qualitative research: credibility, transferability, confirmability and dependability (described in section 2.8.2).

One of the advantages of the focus group method was in the potential for group interaction in the shared construction of experiences of raising a child with CHD. Additionally the asynchronous online format of the groups increased convenience and

ease of participation for geographically distant parents, with ongoing caring duties. It was hoped that this would improve recruitment and participation. The challenges in this research method are the reduced amount of researcher control and influence in the discussion than in face-to-face groups and the influence of group processes on findings. That is to say that group members exert an influence on each other and what happens or what is said in one group might not be replicated in another group, challenging the dependability of the findings.

This study did not make use of participant validation methods on the results of the thematic analysis to check credibility of the findings. It could be argued that this would have improved the credibility of the results of the study. However adding additional participant checks of the themes would have increased the demands of the study on parent participants. Additionally the reality of the length of time between data collection and completion of analysis may have reduced the likelihood of parents' participation, to the detriment of the quality of the validation exercise. Barbour (2001) is critical of the prescriptive use of "technical fixes" such as participant validation as a check for validity. She argues that whilst these methods can be invaluable, for example in participatory action research, use of such methods do not bestow rigour on their own and cannot replace systematic and careful consideration of the qualitative work and its assumptions. Attempts to maintain credibility of the findings have been made by staying close to the raw material during analysis and using direct quotes.

4.3.1.3.1 Qualitative analysis

The clear and systematic analysis procedures outlined by (Braun & Clarke, 2006) were used to analyse the data. Whilst other methods of analysing focus group studies are

available, such as constant comparative analysis and conversational analysis, some of these techniques derive from specific epistemological positions not compatible with a mixed methods methodology. Additionally the lack of non-verbal data would have reduced the effectiveness of more conversationally based analysis techniques.

A weakness of the study might be considered the fact that no measures of inter-rater reliability were presented. Such “quality control” measures (Greenhalgh & Taylor, 1997) might be thought to demonstrate the confirmability of qualitative findings, and the absence of researcher subjectivity in the analysis. However this argument can be thought to derive from a post-positivist position which suggests an objective researcher should seek to find *the* truth which should be the same as that arrived at by any other researcher. An alternative position to this, which this study maintains is aligned to a relativist position that a researcher’s filters cannot be removed but instead should be recognised and acknowledged. The purpose of inter-rater comparisons becomes to add transparency and allow the researchers to reflectively check and widen the coding frame to include alternative ways of coding.

4.3.1.3.2 Researcher bias and reflectivity

One of the common cited criticisms of qualitative research is that it is biased and not reproducible (Mays & Pope, 1995). To maintain the confirmability and dependability of the study findings I used reflective memos throughout coding and analysis (see Appendix M) and I maintained a reflective research journal to increase my reflective awareness of my personal lenses whilst conducting this study. Some of these lenses are described in section 4.6. I have grounded the results in the data and used participants own language to maintain the transparency, and credibility of the current study. Additionally I

met with research supervisors to discuss the themes and analysis and a small amount of transcript was independently coded by multiple coders to refine coding in a reflective manner. I have also considered my own positioning and personal attributes and how these relate to the phenomena of interest in this study (see section 4.6).

4.4 Clinical Implications

Some of the clinical implications of this study have already been described in the chapter. The current findings from this self-selecting sample of parents of children with CHD suggest that while some parents experience high levels of distress that would meet thresholds for PTSD using the DSM-5 criteria this distress might be more fully understood in the context of overwhelming emotional experience of the threat to the life of the child and the need to continue to carry out the work of parenting a child with CHD.

This study found that parents described feeling that others did not understand their experiences and this was related to some parents' desires for peer support. However other parents described the challenges of peer support and difficulties reflecting on their experiences more generally. Avoidance of reflecting on experiences by focussing on the now or putting on a brave face as described in this study may reflect an adaptive coping style and may also be a barrier to seeking help and engaging with services. In the context of high levels of distress manifest in reports of traumatic stress symptoms, some parents may need support to reflect on their experiences.

Whilst a large majority of parents in this study reported positive changes related to their experience of their child's CHD, not all parents did and some parents reported negative changes as a consequence of their experiences. Additionally, given the ongoing

debates about the nature of PTG, and the clinical utility of the construct, Park & Helgeson, (2006) urge for caution in clinical application of interventions designed to increase PTG. Thus clinicians working with parents of children with CHD need to be aware that whilst some parents can and do make positive meaning from their difficult experiences, the picture of changes parents perceive in their lives is likely to be more complicated than current models of PTG allow for.

This study also highlighted an interesting area for clinicians to consider in understanding how tension between the roles of healthcare professional and parent impacts on parents' experiences. It might be easy to assume that healthcare professional parents would be in a better position than other parents however parents in this study described pre-existing healthcare knowledge as both helpful and unhelpful.

4.5 Future Research

This study found high levels of traumatic stress symptoms in a self selecting sample of parents of children with CHD. Further studies investigating prevalence of PTSD in representative clinical samples ensuring good follow-up would be able to offer estimates of prevalence. However as the experiences of parents with children with CHD may be better accounted for by the PMTS and the ADD models of posttraumatic responses, this research would have to consider carefully the definition of the criterion A stressor and the related issues with establishing time since trauma and severity of trauma for this group.

This study found that some parents, similar to other parents of children with serious health conditions, reported a desire for peer support. However peer support was

also constructed as challenging and unhelpful by some parents. Future research could seek to understand what it is that is perceived as helpful for parents of children with CHD in peer support groups, who finds peer support most helpful, and in what contexts can peer support be helpful. This would help inform how peer support groups are facilitated/ supported specifically and more generally how to provide valuable support for parents.

As described in section 4.3.1.1 the findings from this study apply more to the experience of mothers. Fathers' experiences and those of the wider family impacted by the child's CHD such as grandparents are absent from this study. Given previous research findings of differences in the experiences of mothers and fathers future research could investigate the experience of PTSD, PTG and coping in fathers and grandparents related to their child's/ grandchild's CHD. Such accounts could highlight the overlapping and unique aspects of this experience in order to provide an additional layer of triangulation, and another perspective to inform the development of family focussed interventions.

Other possibilities for future research are related to the research methods used in this study. Whilst some parents found the small discussion groups helpful other parents did not and there are limitations in the data due to the focus group methodology. Individual face to face interviews, whilst more resource intensive and more demanding on participants, may reveal different aspects of the experience of parenting a child with CHD not elucidated here.

4.6 Personal Reflections on the research process

During the course of my training and in conducting this study I have changed in many ways, both personally and professionally. Professionally my knowledge of and

skills in research processes, research methods and clinical practice have developed. On a personal level, most obvious is my own journey to motherhood as we welcomed the birth of our son last year. This has been a monumental life change for me, which has inevitably influenced the work as my position and perspective has altered from when first drafts of the study proposal were developed. Going through the early stages of my own pregnancy during the analysis and writing up stages of the study I noticed a different frame of reference became available to me, as a mother to be, anticipating the birth of my own child, in addition to the frames of: young female, researcher, and trainee clinical psychologist. Empathising from this new frame of reference offered a more tangible and imminent application. I was touched by the experiences parents described and the openness with which these experiences were shared. Perhaps I also felt closer to the mothers in the study as I anticipated joining that “club of motherhood”. I kept a reflective journal throughout my training and noted that these feelings of transition and joining were stronger towards the end of my pregnancy compared with the early stages. Returning from maternity leave to write up this thesis I have brought with me new experiences of being a parent with a healthy infant and experiences with networks of other first-time mothers and my own experience of sharing difficulties and emotions with them. These have again influenced my frames through which the study findings are filtered.

I found it particularly difficult to integrate the two methods in the study and I wonder if choosing mixed methods was in part a choice made because of my familiarity and confidence with quantitative methods. I wondered also if this is reinforced by a tendency, in my view, for structures, in our society which award resources and thus confer power, to be swayed by quantitative research. This is reflected in the hierarchies of

research evidence I was taught about in my psychiatric research methods classes which prioritise evidence from meta-analysis and randomised controlled trials.

Working in a paediatric ward in a hospital setting whilst working on this thesis has influenced my practise as my eyes and ears had been opened to the stories from participant parents. I have noticed how I paid more attention than I had in the past to parents experiences, impact on the family and how these in turn impacts on children and young people since conducting this study. Additionally while thinking explicitly about coping strategies with parents and children was very much already a part of my clinical role and I was also conscious of my increased attention and interest in this because of my research interests.

4.7 Conclusions

In conclusion this study found that a small proportion of parents in the self-selecting sample scored above clinical cut offs for PTSD and most parents reported positive changes associated with their child's CHD. However, the key findings from this study highlight the challenges in adopting models of traumatic stress responses that proposes a single triggering traumatic event to understand this experience. Parents of children with CHD, like many other parents of children with a serious health condition, described an overwhelming emotional experience in raising a child with CHD that has irrevocably changed them and their families.

Clinicians should be aware that the ways parents described managing their experiences may also be barriers to seeking help and some parents may need support to reflect on their experiences. This may help make these extraordinary and difficult

experiences, described as incomprehensible to oneself and others, become more understandable.

References

- Abraído-Lanza, A. F., Guier, C., & Colón, R. M. (1998). Psychological Thriving Among Latinas With Chronic Illness. *Journal of Social Issues, 54*(2), 405-424. doi: 10.1111/j.1540-4560.1998.tb01227.x
- Aite, L., Zaccara, A., Mirante, N., Nahom, A., Trucchi, A., Capolupo, I., & Bagolan, P. (2011). Antenatal diagnosis of congenital anomaly: a really traumatic experience? *Journal of Perinatology, 31*(12), 760-763. doi: 10.1038/jp.2011.22
- Ambert, A.-M. (1994). An International Perspective on Parenting: Social Change and Social Constructs. *Journal of Marriage and Family, 56*(3), 529-543. doi: 10.2307/352865
- American Psychiatric Association (APA). (2000). *Diagnostic and statistical manual of mental disorders: DSM-IV* (4th edition ed.). Washington, DC: Author.
- American Psychiatric Association. (2013). *Diagnostic and statistical manual of mental disorders: DSM-5* (5th edition ed.). Washington, DC: Author.
- Angen, M. J. (2000). Evaluating interpretive inquiry: reviewing the validity debate and opening the dialogue. *Qualitative Health Research, 10*(3), 378-395. doi: 10.1177/104973230001000308
- Asch, S. E. (1951). Effects of group pressure upon the modification and distortion of judgments *Groups, leadership and men; research in human relations*. (pp. 177-190). Oxford, England: Carnegie Press.
- Balluffi, A., Kassam-Adams, N., Kazak, A., Tucker, M., Dominguez, T., & Helfaer, M. (2004). Traumatic stress in parents of children admitted to the pediatric intensive care unit. *Pediatric Critical Care Medicine, 5*(6), 547-553. doi: 10.1097/01.pcc.0000137354.19807.44
- Bally, J. M. G., Smith, N. R., Holtslander, L., Duncan, V., Hodgson-Viden, H., Mpofu, C., & Zimmer, M. (2018). A Metasynthesis: Uncovering What Is Known About the Experiences of Families With Children Who Have Life-limiting and Life-threatening Illnesses. *Journal of Pediatric Nursing, 38*, 88-98. doi: 10.1016/j.pedn.2017.11.004
- Barbour, R. S. (2001). Checklists for improving rigour in qualitative research: a case of the tail wagging the dog? *BMJ, 322*(7294), 1115-1117. doi: 10.1136/bmj.322.7294.1115
- Bentall, R. (2004). *Madness explained*. London: Penguin.

- Bevilacqua, F., Palatta, S., Mirante, N., Cuttini, M., Seganti, G., Dotta, A., & Piersigilli, F. (2013). Birth of a child with congenital heart disease: emotional reactions of mothers and fathers according to time of diagnosis. *The Journal of Maternal-Fetal & Neonatal Medicine*, 26(12), 1249-1253.
- Bibring, E. (1943) The Conception of the Repetition Compulsion, *The Psychoanalytic Quarterly*, 12:4, 486-519, DOI: [10.1080/21674086.1943.11925548](https://doi.org/10.1080/21674086.1943.11925548)
- Biesta, G. (2010). *SAGE Handbook of Mixed Methods in Social & Behavioral Research* (2 ed.). Thousand Oaks, California: SAGE Publications, Inc.
- Bifulco, A., Figueiredo, B., Guedeney, N., Gorman, L. L., Hayes, S., Muzik, M., . . . Henshaw, C. A. (2004). Maternal attachment style and depression associated with childbirth: preliminary results from a European and US cross-cultural study. *British Journal of Psychiatry. Supplement*, 46, s31-37.
- Blevins, C. A., Weathers, F. W., Davis, M. T., Witte, T. K., & Domino, J. L. (2015). The Posttraumatic Stress Disorder Checklist for DSM-5 (PCL-5): Development and Initial Psychometric Evaluation. *Journal of Traumatic Stress*, 28(6), 489-498. doi: [10.1002/jts.22059](https://doi.org/10.1002/jts.22059)
- Bonanno, G. A. (2013). Meaning making, adversity, and regulatory flexibility. *Memory*, 21(1), 150-156. doi: [10.1080/09658211.2012.745572](https://doi.org/10.1080/09658211.2012.745572)
- Bonanno, G. A., Pat-Horenczyk, R., & Noll, J. (2011). Coping flexibility and trauma: The Perceived Ability to Cope With Trauma (PACT) scale. *Psychological Trauma: Theory, Research, Practice, and Policy*, 3(2), 117-129. doi: [10.1037/a0020921](https://doi.org/10.1037/a0020921)
- Bornstein, M. H. (2006) Parenting science and practice. In: Damon W., Renninger K. A., Sigel I. E., Eds. *Handbook of child psychology: Vol. 4. Child psychology in practice*. 6th Ed. New York: Wiley; 2006. pp. 893–949.
- Bosquet Enlow, M., Egeland, B., Carlson, E., Blood, E., & Wright, R. J. (2014). Mother-infant attachment and the intergenerational transmission of posttraumatic stress disorder. *Development and Psychopathology*, 26(1), 41-65. doi: [10.1017/s0954579413000515](https://doi.org/10.1017/s0954579413000515)
- Bowlby, J. (1969). *Attachment and Loss: Vol 1: Attachment*. London: Hogarth Press.
- Bracken, P., Thomas, P., Timimi, S., Asen, E., Behr, G., Beuster, C., . . . Yeomans, D. (2012). Psychiatry beyond the current paradigm. *British Journal of Psychiatry*, 201, 430-434. Doi: [10.1192/bjp.bp112.109447](https://doi.org/10.1192/bjp.bp112.109447).
- Bratt, E. L., Jarvholm, S., Ekman-Joelsson, B. M., Mattson, L. A., & Mellander, M. (2015). Parent's experiences of counselling and their need for support following a

- prenatal diagnosis of congenital heart disease--a qualitative study in a Swedish context. *BMC Pregnancy and Childbirth*, 15, 171. doi: 10.1186/s12884-015-0610-4
- Braun, V., & Clarke, V. (2006). Using thematic analysis in psychology. *Qualitative Research in Psychology*, 3(2), 77-101. doi: 10.1191/1478088706qp063oa
- Braun, V., & Clarke, V. (2013). *Successful qualitative research: a practical guide for beginners*. London: Sage.
- Brosig, C. L., Whitstone, B. N., Frommelt, M. A., Frisbee, S. J., & Leuthner, S. R. (2007). Psychological distress in parents of children with severe congenital heart disease: the impact of prenatal versus postnatal diagnosis. *Journal of Perinatology*, 27(11), 687-692. doi: 10.1038/sj.jp.7211807
- Brown, K. L., Wray, J., Knowles, R. L., Crowe, S., Tregay, J., Ridout, D., . . . Bull, C. (2016). Infant deaths in the UK community following successful cardiac surgery: building the evidence base for optimal surveillance, a mixed-methods study. *Health Services and Delivery Research*. doi: 10.3310/hsdr04190
- Brown, K. M., Elliott, S. J., Leatherdale, S. T., & Robertson-Wilson, J. (2015). Searching for rigour in the reporting of mixed methods population health research: a methodological review. *Health Education Research*, 30(6), 811-839. doi: 10.1093/her/cyv046
- British Psychological Society. (2010). Code of Human Research Ethics. Leicester: British Psychological Society.
- Bryman, A. (2008). *The SAGE Handbook of Social Research Methods*. London: SAGE Publications Ltd.
- Burke, K., Muscara, F., McCarthy, M., Dimovski, A., Hearps, S., Anderson, V., & Walser, R. (2014). Adapting acceptance and commitment therapy for parents of children with life-threatening illness: pilot study. *Fam Syst Health*, 32(1), 122-127. doi: 10.1037/fsh0000012
- Burton, C. L., Yan, O. H., Pat-Horenczyk, R., Chan, I. S., Ho, S., & Bonanno, G. A. (2012). Coping flexibility and complicated grief: a comparison of American and Chinese samples. *Depression and Anxiety*, 29(1), 16-22. doi: 10.1002/da.20888
- Cahill, S. P., & Pontoski, K. (2005). Posttraumatic Stress Disorder and Acute Stress Disorder I: Their Nature and Assessment Considerations. *Psychiatry (Edgmont)*, 2(4), 14-25.

- Calhoun, L. G., & Tedeschi, R. (2001). *Posttraumatic growth: The positive lessons of loss*.
- Carey, M. A. (1993). The group effect in focus groups: Planning, implementing and interpreting focus group research. In J. Morse (Ed.), *Critical Issues in Qualitative Research Methods* (pp. 225 - 261). Newbury Park, CA: Sage.
- Carlsson, T., Bergman, G., Melander Marttala, U., Wadensten, B., & Mattsson, E. (2015). Information following a diagnosis of congenital heart defect: experiences among parents to prenatally diagnosed children. *PloS One*, *10*(2), e0117995. doi: 10.1371/journal.pone.0117995
- Charmaz, K. (2006). *Constructing Grounded Theory: A practical guide through qualitative analysis*. London, UK.: Sage Publications.
- Cheng, C. (2001). Assessing coping flexibility in real-life and laboratory settings: A multimethod approach. *Journal of Personality and Social Psychology*, *80*(5), 814-833. doi: 10.1037/0022-3514.80.5.814
- Cheng, C., Lau, H.-P. B., & Chan, M.-P. S. (2014). Coping flexibility and psychological adjustment to stressful life changes: A meta-analytic review. *Psychological Bulletin*, *140*(6), 1582-1607. doi: 10.1037/a0037913
- Churchill, R. (2003). Critical appraisal. In M. Prince, R. Stewart, T. Ford & M. Hotopf (Eds.), *Practical Psychiatric Epidemiology* (pp. 291 - 314). Oxford: Oxford University Press.
- Clark, S. M., & Miles, M. S. (1999). Conflicting responses: the experiences of fathers of infants diagnosed with severe congenital heart disease. *Journal for Specialists in Pediatric Nursing*, *4*(1), 7-14.
- Coffey, J. S. (2006). Parenting a child with chronic illness: a metasynthesis. *Pediatric Nursing*, *32*(1), 51-59.
- Cohen, J. (1992). A power primer. *Psychological Bulletin*, *112*(1), 155-159.
- Collogan, L., Tuma, F., Dolan-Sewell, R., Borja, S., & Fleischman, A. (2004). Ethical Issues Pertaining to Research in the Aftermath of Disaster. *Journal of Traumatic Stress*, *17*(5), 363-372. doi: 10.1023/B:JOTS.0000048949.43570.6a
- Colville, G., & Cream, P. (2009). Posttraumatic growth in parents after a child's admission to intensive care: maybe Nietzsche was right? *Intensive Care Medicine*, *35*(5), 919-923. doi: 10.1007/s00134-009-1444-1

- Critical appraisal skills programme (CASP). (2013). *Qualitative Research check list*. Retrieved on 26th November 2013 from <http://www.casp-uk.net/#!casp-tools-checklists/c18f8>
- Das, S., & Kramer, A. (2013). *Self-censorship on facebook*.
- Davis, C. C., Brown, R. T., Bakeman, R., & Campbell, R. (1998). Psychological adaptation and adjustment of mothers of children with congenital heart disease: stress, coping, and family functioning. *Journal of Pediatric Psychology*, 23(4), 219-228.
- Division of Clinical Psychology, (2013). *Classification of behaviour and experiences in relation to functional diagnosis: Time for a paradigm shift*. Leicester, England: British Psychological Society.
- Doherty, N., McCusker, C. G., Molloy, B., Mulholland, C., Rooney, N., Craig, B., . . . Casey, F. (2009). Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease. *Journal of Reproductive and Infant Psychology*, 27(4), 390-400. doi: 10.1080/02646830903190920
- Dolk, H., Loane, M., & Garne, E. (2011). Congenital heart defects in Europe: prevalence and perinatal mortality, 2000 to 2005. *Circulation*, 123(8), 841-849. doi: 10.1161/circulationaha.110.958405
- Eiser, C. (1993). *Growing up with a chronic disease: The impact on children and their families*. London: Jessica Kingsley Publishers Ltd.
- Feinstein, J. A., Benson, D. W., Dubin, A. M., Cohen, M. S., Maxey, D. M., Mahle, W. T., . . . Martin, G. R. (2012). Hypoplastic Left Heart Syndrome: Current Considerations and Expectations. *Journal of the American College of Cardiology*, 59(1, Supplement), S1-S42. doi: <http://dx.doi.org/10.1016/j.jacc.2011.09.022>
- Field, A. (2009). *Discovering statistics using SPSS* (Third ed.). London: SAGE Publishing.
- Fisher, H. R. (2001). The needs of parents with chronically sick children: a literature review. *Journal of Advanced Nursing*, 36(4), 600-607.
- Forinder, U., & Norberg, A. L. (2014). Posttraumatic growth and support among parents whose children have survived stem cell transplantation. *J Child Health Care*, 18(4), 326-335. doi: 10.1177/1367493513496666
- Franck, L. S., Mcquillan, A., Wray, J., Grocott, M. P., & Goldman, A. (2010). Parent stress levels during children's hospital recovery after congenital heart surgery. *Pediatric Cardiology*, 31(7), 961-968.

- Franich-Ray, C., Bright, M. A., Anderson, V., Northam, E., Cochrane, A., Menahem, S., & Jordan, B. (2013). Trauma reactions in mothers and fathers after their infant's cardiac surgery. *Journal of Pediatric Psychology, 38*(5), 494-505. doi: 10.1093/jpepsy/jst015
- Frank, A. W. (1995). *The wounded storyteller: Body illness and ethics*. Chicago: Chicago University Press.
- Frazier, P., Conlon, A., & Glaser, T. (2001). Positive and negative life changes following sexual assault. *Journal of Consulting and Clinical Psychology, 69*(6), 1048-1055.
- Friedman, M. J. (2013). Finalizing PTSD in DSM-5: Getting Here From There and Where to Go Next. *Journal of Traumatic Stress, 26*(5), 548-556. doi: 10.1002/jts.21840
- Friedman, Resick & Keane (2007) PTSD: Twenty-five years of progress and challenges. In M. J. Friedman, T. M. Keane & P. A. Resick (Eds.) *Handbook of PTSD: Science and practice* (pp3-18). New York, NY: Guildford Press
- Frissa, S., Hatch, S. L., Gazard, B., Fear, N. T., & Hotopf, M. (2013). Trauma and current symptoms of PTSD in a South East London community. *Social Psychiatry and Psychiatric Epidemiology, 48*(8), 1199-1209. doi: 10.1007/s00127-013-0689-8
- George, A., Vickers, M. H., Wilkes, L., & Barton, B. (2007). Chronic grief: Experiences of working parents of children with chronic illness. *Contemporary Nurse, 23*(2), 228-242. doi: 10.5172/conu.2006.23.2.228
- Gravelle, A. M. (1997). Caring for a child with a progressive illness during the complex chronic phase: parents' experience of facing adversity. *Journal of Advanced Nursing, 25*(4), 738-745.
- Greenhalgh, T., & Taylor, R. (1997). How to read a paper: Papers that go beyond numbers (qualitative research). *BMJ, 315*(7110), 740-743. doi: 10.1136/bmj.315.7110.740
- Grotstein, J. S. (1981). *Splitting and projective identification*. New York: Aronson.
- Guba, E. G., & Lincoln, Y. S. (1994). Competing paradigms in qualitative research. In N. K. Denzin & Y. S. Lincoln (Eds.), *Handbook of qualitative research* (pp. 105-117). London: Sage.
- Hall, R. (2013). Mixed methods: In search of a paradigm. In T. Le & Q. Le (Eds.), *Conducting research in a changing and challenging world* (pp. 71-78): Nova Science Publishers.
- Harvey, A. G., & Bryant, R. A. (2002). Acute stress disorder: a synthesis and critique. *Psychological Bulletin, 128*(6), 886-902.

- Harvey, K. A., Kovalesky, A., Woods, R. K., & Loan, L. A. (2013). Experiences of mothers of infants with congenital heart disease before, during, and after complex cardiac surgery. *Heart and Lung, 42*(6), 399-406. doi: 10.1016/j.hrtlng.2013.08.009
- Heath, G., Farre, A. & Shaw, K. (2017) Parenting a child with chronic illness as they transition into adulthood: A systematic review and thematic synthesis of parents' experiences. *Patient Education and Counselling, 100* (1), 76-92. doi: 10.1016/j.pec.2016.08.011
- Helfricht, S., Landolt, M. A., Moergeli, H., Hepp, U., Wegener, D., & Schnyder, U. (2009). Psychometric evaluation and validation of the German version of the Acute Stress Disorder Scale across two distinct trauma populations. *Journal of Traumatic Stress, 22*(5), 476-480. doi: 10.1002/jts.20445
- Helfricht, S., Latal, B., Fischer, J. E., Tomaske, M., & Landolt, M. A. (2008). Surgery-related posttraumatic stress disorder in parents of children undergoing cardiopulmonary bypass surgery: a prospective cohort study. *Pediatric Critical Care Medicine, 9*(2), 217-223. doi: 10.1097/PCC.0b013e318166eec3
- Helgeson, V. S., Reynolds, K. A., & Tomich, P. L. (2006). A meta-analytic review of benefit finding and growth. *Journal of Consulting and Clinical Psychology, 74*(5), 797-816. doi: 10.1037/0022-006X.74.5.797
- Hilton-Kamm, D., Sklansky, M., & Chang, R.-K. (2014). How not to tell parents about their child's new diagnosis of congenital heart disease: an internet survey of 841 parents. *Pediatric Cardiology, 35*(2), 239-252.
- Horsch, A., Brooks, C., & Fletcher, H. (2013). Maternal coping, appraisals and adjustment following diagnosis of fetal anomaly. *Prenatal Diagnosis, 33*(12), 1137-1145. doi: 10.1002/pd.4207
- Hungerbuehler, I., Vollrath, M. E., & Landolt, M. A. (2011). Posttraumatic growth in mothers and fathers of children with severe illnesses. *Journal of Health Psychology, 16*(8), 1259-1267. doi: 10.1177/1359105311405872
- Huxter, W. & Holden, J. (2015, October 7). The new Congenital Heart Disease review: 43rd update. [Blog post]. Retrieved from <https://www.england.nhs.uk/2015/10/john-holden-43/>
- IBM Corp. (2010). IBM SPSS Statistics for Windows (Version 19.0). Armonk, NY: IBM Corp.

- Ikemba, C. M., Kozinetz, C. A., Feltes, T. F., Fraser, C. D., Jr., McKenzie, E. D., Shah, N., & Mott, A. R. (2002). Internet use in families with children requiring cardiac surgery for congenital heart disease. *Pediatrics*, *109*(3), 419-422.
- Jackson, A. C., Frydenberg, E., Liang, R. P. T., Higgins, R. O., & Murphy, B. M. (2015). Familial impact and coping with child heart disease: a systematic review. *Pediatric Cardiology*, *36*(4), 695-712. doi: 10.1007/s00246-015-1121-9
- Johnson, R. B., & Onwuegbuzie, A. J. (2004). Mixed Methods Research: A Research Paradigm Whose Time Has Come. *Educational Researcher*, *33*(7), 14-26.
- Johnson, R. B., Onwuegbuzie, A. J., & Turner, L. A. (2007). Toward a Definition of Mixed Methods Research. *Journal of Mixed Methods Research*, *1*(2), 112-133. doi: 10.1177/1558689806298224
- Johnstone, L. (2014). *A straight-talking introduction to psychiatric diagnosis*. Ross-on-Wye, England: PCCS Books.
- Johnstone, L. (2018). Psychological formulation as an alternative to psychiatric diagnosis. *Journal of Humanistic Psychology* *58* (1), 30-46. Doi: 10.1177/0022167817722230
- Joseph, S., Alex Linley, P., & Harris, G. J. (2004). Understanding positive change following trauma and adversity: Structural clarification. *Journal of Loss and Trauma*, *10*(1), 83-96. doi: 10.1080/15325020490890741
- Joseph, S., & Linley, P. A. (2008). Positive Psychological Perspectives on Posttraumatic Stress: An Integrative Psychosocial Framework In S. Joseph & P. A. Linley (Eds.), *Trauma, Recovery, and Growth: Positive Psychological Perspectives on Posttraumatic Stress* (pp. 3-20). New Jersey: John Wiley & Sons.
- Joseph, S., Maltby, J., Wood, A. M., Stockton, H., Hunt, N., & Regel, S. (2012). The Psychological Well-Being—Posttraumatic Changes Questionnaire (PWB-PTCQ): Reliability and validity. *Psychological Trauma: Theory, Research, Practice, and Policy*, *4*(4), 420-428. doi: 10.1037/a0024740
- Joseph, S., & Williams, R. (2005). Understanding Posttraumatic Stress: Theory, Reflections, Context and Future. *Behavioural and Cognitive Psychotherapy*, *33*(4), 423-441. doi: 10.1017/S1352465805002328
- Kahneman, D. (1973). *Attention and Effort*, New Jersey: Prentice-Hall.
- Kaisar, J., Strodl, E., Schweitzer, R. D., & Radford, D. (2012). A qualitative study of adversity activated development and resilience in young people with CHD and their parents. In K. Gow & M. J. Celinski (Eds.), *Individual trauma: recovering*

- from deep wounds and exploring the potential for renewal* (pp. 221-237). New York: Nova.
- Kassam-Adams, N. (2006). Introduction to the special issue: Posttraumatic stress related to pediatric illness and injury. *Journal of Pediatric Psychology, 31*(4), 337-342. doi: 10.1093/jpepsy/jsj052
- Kazak, A. E., Barakat, L. P., Meeske, K., Christakis, D., Meadows, A. T., Casey, R., . . . Stuber, M. L. (1997). Posttraumatic stress, family functioning, and social support in survivors of childhood leukemia and their mothers and fathers. *Journal of Consulting and Clinical Psychology, 65*(1), 120-129.
- Kazak, A. E., Kassam-Adams, N., Schneider, S., Zelikovsky, N., Alderfer, M. A., & Rourke, M. (2006). An integrative model of pediatric medical traumatic stress. *Journal of Pediatric Psychology, 31*(4), 343-355. doi: 10.1093/jpepsy/jsj054
- Kepreotes, E., Keatinge, D. and Stone, T. (2010), The experience of parenting children with chronic health conditions: a new reality. *Journal of Nursing and Healthcare of Chronic Illness, 2*: 51–62. doi:10.1111/j.1752-9824.2010.01047.x
- Kessler, R. C., Sonnega, A., Bromet, E., Hughes, M., & Nelson, C. B. (1995). Posttraumatic stress disorder in the National Comorbidity Survey. *Archives of General Psychiatry, 52*(12), 1048-1060.
- Kirk, S. A. & Kutchins, H. (1997). *Making us crazy: DSM The psychiatric bible and the creation of mental disorder*. New York, NY: Free Press.
- Klass, P. (1987). *A not entirely benign procedure: Four years as a medical student*. New York: Penguin.
- Kosta, L., Harms, L., Franich-Ray, C., Anderson, V., Northam, E., Cochrane, A., Menaham, S., & Jordan, B. (2015). Parental experiences of their infant's hospitalization for cardiac surgery. *Child: Care, Health and Development, 41*(6), 1057-1065.
- Kovacs, M., Finkelstein, R., Feinberg, T. L., Crouse-Novak, M., Paulauskas, S., & Pollock, M. (1985). Initial Psychologic Responses of Parents to the Diagnosis of Insulin-dependent Diabetes Mellitus in Their Children. *Diabetes Care, 8*(6), 568-575. doi: 10.2337/diacare.8.6.568
- Kramish Campbell, M., Meier, A., Carr, C., Enga, Z., James, A. S., Reedy, J., & Zheng, B. (2001). Health behavior changes after colon cancer: a comparison of findings from face-to-face and on-line focus groups. *Family and Community Health, 24*(3), 88-103.

- Krueger, R. A., & Casey, M. A. (2009). *Focus Groups: A practical guide for applied research* (4th ed.). Los Angeles: SAGE.
- Kuhn, T. S. (1970). *The structure of scientific revolutions*. Chicago: University of Chicago Press.
- Landolt, M. A., Buechel, E. V., & Latal, B. (2011). Predictors of parental quality of life after child open heart surgery: a 6-month prospective study. *Journal of Pediatrics*, *158*(1), 37-43. doi: 10.1016/j.jpeds.2010.06.037
- Landolt, M. A., Ystrom, E., Sennhauser, F. H., Gnehm, H. E., & Vollrath, M. E. (2012). The mutual prospective influence of child and parental posttraumatic stress symptoms in pediatric patients. *Journal of Child Psychology and Psychiatry and Allied Disciplines*, *53*(7), 767-774. doi: 10.1111/j.1469-7610.2011.02520.x
- Landolt, M. A., Ystrom, E., Stene-Larsen, K., Holmstrom, H., & Vollrath, M. E. (2014). Exploring causal pathways of child behavior and maternal mental health in families with a child with congenital heart disease: a longitudinal study. *Psychological Medicine*, *44*(16), 3421-3433. doi: 10.1017/s0033291713002894
- Lazarus, R. S., & Folkman, S. (1984). *Stress, appraisal and coping*. New York: Springer.
- Lefkowitz, D. S., Baxt, C., & Evans, J. R. (2010). Prevalence and Correlates of Posttraumatic Stress and Postpartum Depression in Parents of Infants in the Neonatal Intensive Care Unit (NICU). *Journal of Clinical Psychology in Medical Settings*, *17*(3), 230-237. doi: 10.1007/s10880-010-9202-7
- Li, Y., Cao, F., Cao, D., Wang, Q., & Cui, N. (2012). Predictors of posttraumatic growth among parents of children undergoing inpatient corrective surgery for congenital disease. *Journal of Pediatric Surgery*, *47*(11), 2011-2021. doi: <http://dx.doi.org/10.1016/j.jpedsurg.2012.07.005>
- Lincoln, Y. S., & Guba, E. G. (1985). *Naturalistic Enquiry*. Newbury Park, CA: Sage.
- Linley, P. A., & Joseph, S. (2004). Positive change following trauma and adversity: a review. *Journal of Traumatic Stress*, *17*(1), 11-21. doi: 10.1023/B:JOTS.0000014671.27856.7e
- Lok, S. W., & Menahem, S. (2004). Parental perception of small ventricular septal defects in childhood. *Journal of Paediatrics and Child Health*, *40*(4), 180-183.
- Lowes, L. and Lyne, P. (2000), Chronic sorrow in parents of children with newly diagnosed diabetes: a review of the literature and discussion of the implications for nursing practice. *Journal of Advanced Nursing*, *32*: 41-48. doi:10.1046/j.1365-2648.2000.01418.x

- MacDonald, H. and Callery, P. (2008). Parenting children requiring complex care: a journey through time. *Child: Care, Health and Development*, 34: 207–213. doi:10.1111/j.1365-2214.2007.00790.x
- Maltby, H. J., Kristjanson, L. and 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. doi:10.1046/j.1440-172X.2003.00445.x
- Masse, R. (2000). Qualitative and quantitative analyses of psychological distress: methodological complementarity and ontological incommensurability. *Qualitative Health Research*, 10(3), 411-423. doi: 10.1177/104973200129118426
- Maxwell, J. A., & Mittapalli, K. (2010). *SAGE Handbook of Mixed Methods in Social & Behavioral Research* (2 ed.). Thousand Oaks, California: SAGE Publications, Inc.
- Maxwell, J. A. (2004). Causal Explanation, Qualitative Research, and Scientific Inquiry in Education. *Educational Researcher*, 33(2), 3-11. doi: 10.3102/0013189x033002003
- Mays, N., & Pope, C. (1995). Qualitative Research: Rigour and qualitative research. *BMJ*, 311(6997), 109-112. doi: 10.1136/bmj.311.6997.109
- Mays, N., & Pope, C. (2000). Assessing quality in qualitative research. *BMJ : British Medical Journal*, 320(7226), 50-52.
- McManus S, B. P., Jenkins R, Brugha T. (eds.). (2016). *Mental health and wellbeing in England: Adult Psychiatric Morbidity Survey 2014*. Leeds: NHS Digital.
- McMillen, J. C., & Fisher, R. H. (1998). The Perceived Benefit Scales: Measuring perceived positive life changes after negative events. *Social Work Research*, 22(3), 173-187. doi: 10.1093/swr/22.3.173
- Messias, D. K., Gilliss, C. L., Sparacino, P. S., Tong, E. M., & Foote, D. (1995). Stories of transition: Parents recall the diagnosis of congenital heart defect. *Family Systems Medicine*, 13(3-4), 367.
- Mishel, M. H. (1988), Uncertainty in Illness. *Image: the Journal of Nursing Scholarship*, 20: 225–232. doi:10.1111/j.1547-5069.1988.tb00082.x
- Moola, F. J. (2012). "This is the best fatal illness that you can have": contrasting and comparing the experiences of parenting youth with cystic fibrosis and congenital heart disease. *Qualitative Health Research*, 22(2), 212-225. doi: 10.1177/1049732311421486

- Morgan, D. L. (1998). Practical strategies for combining qualitative and quantitative methods: applications to health research. *Qualitative Health Research*, 8(3), 362-376.
- Morgan, D. L. (2007). Paradigms Lost and Pragmatism Regained. *Journal of Mixed Methods Research*, 1(1), 48-76. doi: 10.1177/2345678906292462
- Morgan, D. L. (2014). *Integrating qualitative and quantitative methods. A pragmatic approach*. London: SAGE Publications Ltd.
- Murray Parkes, C., & Prigerson, H. (2010). *Bereavement; Studies of Grief in Adult Life*. London: Penguin.
- Mussatto, K. A., Hoffmann, R. G., Hoffman, G. M., Tweddell, J. S., Bear, L., Cao, Y., & Brosig, C. (2014). Risk and Prevalence of Developmental Delay in Young Children With Congenital Heart Disease. *Pediatrics*, 133(3), e570-e577. doi: 10.1542/peds.2013-2309
- Newman, E., Risch, E., & Kassam-Adams, N. (2006). Ethical issues in trauma-related research: a review. *J Empir Res Hum Res Ethics*, 1(3), 29-46. doi: 10.1525/jer.2006.1.3.29
- NHS England Board. (2015). New Congenital Heart Disease Review, : NHS England.
- NHS Health Research Authority (2013) Do I need NHS REC approval? Decision Tool retrieved from <http://www.hra-decisiontools.org.uk/ethics/>
- Nugent, N. R., Ostrowski, S., Christopher, N. C., & Delahanty, D. L. (2007). Parental posttraumatic stress symptoms as a moderator of child's acute biological response and subsequent posttraumatic stress symptoms in pediatric injury patients. *Journal of Pediatric Psychology*, 32(3), 309-318. doi: 10.1093/jpepsy/jsl005
- Office for National Statistics (ONS). (2016). Internet access – households and individuals: 2016: ONS.
- Onwuegbuzie, A. J., Dickinson, W. B., Leech, N. L., & Zoran, A. G. (2009). A Qualitative Framework for Collecting and Analyzing Data in Focus Group Research. *International Journal of Qualitative Methods*, 8(3), 1-21. doi: 10.1177/160940690900800301
- Papadopoulos, R. K. (2007). Refugees, trauma and Adversity-Activated Development. *European Journal of Psychotherapy & Counselling*, 9(3), 301-312. doi: 10.1080/13642530701496930
- Park, C. L., & Helgeson, V. S. (2006). Introduction to the special section: Growth following highly stressful life events--Current status and future directions. *Journal*

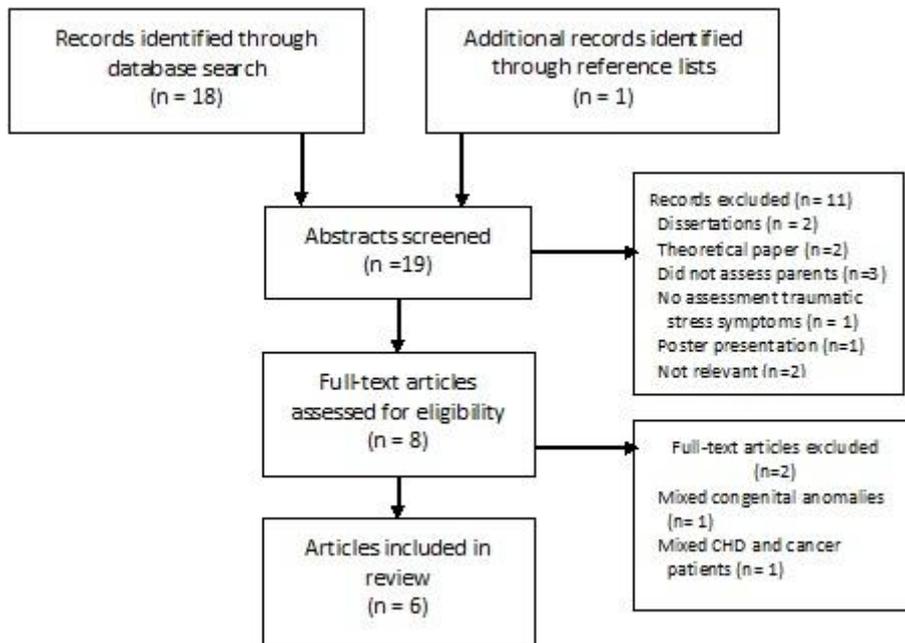
- of Consulting and Clinical Psychology*, 74(5), 791-796. doi: 10.1037/0022-006X.74.5.791
- Parker, R. (2005). *Torn in two: The experience of maternal ambivalence*. London: Virago
- Petersen, S., Peto, V., & Rayner, M. (2003). Congenital heart disease statistics: British Heart Foundation Health Promotion Research Group.
- Picoraro, J. A., Womer, J. W., Kazak, A. E., & Feudtner, C. (2014). Posttraumatic growth in parents and pediatric patients. *Journal of Palliative Medicine*, 17(2), 209-218. doi: 10.1089/jpm.2013.0280
- Porter, S. (2007). Validity, trustworthiness and rigour: reasserting realism in qualitative research. *Journal of Advanced Nursing*, 60(1), 79-86. doi: 10.1111/j.1365-2648.2007.04360.x
- Prince, M. (2003). Cross-sectional surveys. In M. Prince, R. Stewart, T. Ford & M. Hotopf (Eds.), *Practical Psychiatric Epidemiology* (pp. 111 - 129). Oxford: Oxford University Press.
- Qualtrics. (2015). Qualtrics Online Survey Platform (Version 2015). Utah, USA: Available from <http://www.qualtrics.com>.
- Ravindran, V. P., & Rempel, G. R. (2011). Grandparents and siblings of children with congenital heart disease. *Journal of Advanced Nursing*, 67(1), 169-175.
- Re, J., Dean, S., & Menahem, S. (2013). Infant cardiac surgery: mothers tell their story: a therapeutic experience. *World J Pediatr Congenit Heart Surg*, 4(3), 278-285. doi: 10.1177/2150135113481480
- Rempel, G. R., & Harrison, M. J. (2007). Safeguarding precarious survival: parenting children who have life-threatening heart disease. *Qualitative Health Research*, 17(6), 824-837.
- Rempel, G. R., Ravindran, V., Rogers, L. G., & Magill-Evans, J. (2013). Parenting under Pressure: a grounded theory of parenting young children with life-threatening congenital heart disease. *Journal of Advanced Nursing*, 69(3), 619-630.
- Rempel, G. R., Rogers, L. G., Ravindran, V., & Magill-Evans, J. (2012). Facets of parenting a child with hypoplastic left heart syndrome. *Nursing Research and Practice*, 2012.
- Rini, C., Manne, S., DuHamel, K. N., Austin, J., Ostroff, J., Boulad, F., . . . Redd, W. H. (2004). Mothers' perceptions of benefit following pediatric stem cell transplantation: a longitudinal investigation of the roles of optimism, medical risk,

- and sociodemographic resources. *Annals of Behavioral Medicine*, 28(2), 132-141. doi: 10.1207/s15324796abm2802_9
- Robson, C. (2011). *Real World Research* (3rd ed.). West Sussex: John Wiley & Sons.
- Rolfe, G. (2006). Validity, trustworthiness and rigour: quality and the idea of qualitative research. *Journal of Advanced Nursing*, 53(3), 304-310. doi: 10.1111/j.1365-2648.2006.03727.x
- Rolfe, G. (2007). In response to Porter S. Validity, trustworthiness and rigour: reasserting realism in qualitative research. *Journal of Advanced Nursing* 60 (1), 79-86. *Journal of Advanced Nursing*, 60(1), 108-109. doi: 10.1111/j.1365-2648.2007.04366.x
- Rychik, J., Donaghue, D. D., Levy, S., Fajardo, C., Combs, J., Zhang, X., . . . Diamond, G. S. (2013). Maternal psychological stress after prenatal diagnosis of congenital heart disease. *Journal of Pediatrics*, 162(2), 302-307.e301. doi: 10.1016/j.jpeds.2012.07.023
- Sandelowski, M. & Barroso, J. (2006). *Handbook for synthesizing qualitative research*. New York: Springer
- Sayer, A. (1992). *Method in social science: A realist approach* (2nd ed.). London: Routledge.
- Sayer, A. (2000). *Realism and Social Science*. London: SAGE.
- Schneider, S. J., Kerwin, J., Frechtling, J., & Vivari, B. A. (2002). Characteristics of the Discussion in Online and Face-to-Face Focus Groups. *Social Science Computer Review*, 20(1), 31-42. doi: 10.1177/089443930202000104
- Scorgie, K., & Sobsey, D. (2000). Transformational outcomes associated with parenting children who have disabilities. *Mental Retardation*, 38(3), 195-206. doi: 10.1352/0047-6765(2000)038<0195:TOAWPC>2.0.CO;2
- Shadish, W. R. (2002). Experimental and quasi-experimental designs for generalized causal inference / William R. Shadish, Thomas D. Cook, Donald T. Campbell (pp. 0-0).
- Shafran, R., Thordarson, D. S., & Rachman, S. (1996). Thought-action fusion in obsessive compulsive disorder. *Journal of Anxiety Disorders*, 10(5), 379-391. doi: [http://dx.doi.org/10.1016/0887-6185\(96\)00018-7](http://dx.doi.org/10.1016/0887-6185(96)00018-7)
- Shakespeare-Finch, J., & Lurie-Beck, J. (2014). A meta-analytic clarification of the relationship between posttraumatic growth and symptoms of posttraumatic

- distress disorder. *Journal of Anxiety Disorders*, 28(2), 223-229. doi: <https://doi.org/10.1016/j.janxdis.2013.10.005>
- Shaw, R. J., Deblois, T., Ikuta, L., Ginzburg, K., Fleisher, B., & Koopman, C. (2006). Acute Stress Disorder Among Parents of Infants in the Neonatal Intensive Care Nursery. *Psychosomatics*, 47(3), 206-212. doi: 10.1176/appi.psy.47.3.206
- Shiue, Y.-C., Chiu, C.-M., & Chang, C.-C. (2010). Exploring and mitigating social loafing in online communities. *Computers in Human Behavior*, 26(4), 768-777. doi: <http://dx.doi.org/10.1016/j.chb.2010.01.014>
- Simpson, J. A., & Rholes, W. S. (2004). Anxious attachment and depressive symptoms: an interpersonal perspective. In W. S. Rholes & J. A. Simpson (Ed.), *Adult Attachment: Theory, Research and Clinical Implications*. (pp. 408 - 437). New York: Guildford Press.
- Smith, J. A. (1996). Beyond the divide between cognition and discourse: Using interpretative phenomenological analysis in health psychology. *Psychology & Health*, 11(2), 261-271. doi: 10.1080/08870449608400256
- Smith, J., Cheater, F., & Bekker, H. (2013). Parents' experiences of living with a child with a long-term condition: a rapid structured review of the literature. *Health Expectations*, 18, 452-474. doi:10.1111/hex.12040
- Soulvie, M. A., Desai, P. P., White, C. P., & Sullivan, B. N. (2012). Psychological Distress Experienced by Parents of Young Children With Congenital Heart Defects: A Comprehensive Review of Literature. *Journal of Social Service Research*, 38(4), 484-502. doi: 10.1080/01488376.2012.696410
- Spijkerboer, A. W., Helbing, W. A., Bogers, A. J., Van Domburg, R. T., Verhulst, F. C., & Utens, E. M. (2007). Long-term psychological distress, and styles of coping, in parents of children and adolescents who underwent invasive treatment for congenital cardiac disease. *Cardiology in the Young*, 17(6), 638-645. doi: 10.1017/s1047951107001333
- Springett, A., Budd, J., Draper, E. S., Kurinczuk, J. J., Medina, J., Rankin, J., . . . Morris, J. K. (2014). *Congenital Anomaly Statistics 2012: England and Wales*. London.
- Stein R.E. K. & Jessop, D. J. (1982). A noncategorical approach to chronic childhood illness. *Public Health Reports*, 97, 354-362.
- Stewart, J. L., & Mishel, M. H. (2000). Uncertainty in childhood illness: a synthesis of the parent and child literature. *Scholarly Inquiry for Nursing Practice*, 14(4), 299-319; discussion 321-296.

- Stockton, H., Joseph, S., & Hunt, N. (2014). Expressive writing and posttraumatic growth: An Internet-based study. *Traumatology*, 20 (2). pp. 75-83.
- Stoppelbein, L., & Greening, L. (2007). Brief report: the risk of posttraumatic stress disorder in mothers of children diagnosed with pediatric cancer and type I diabetes. *Journal of Pediatric Psychology*, 32(2), 223-229. doi: 10.1093/jpepsy/jsj120
- Summerfield, D. (2001). The invention of posttraumatic stress disorder and the social usefulness of a psychiatric category. *BMJ*, 322(7278), 95-98. doi:
- Tan, R. & McConvey, R. (2014). Teaching on diagnosis: An interprofessional reflection, *Clinical Psychology Forum*, 261, 22-25.
- Tedeschi, R. G., & Calhoun, L. G. (1995). *Trauma and transformation: Growing in the aftermath of suffering*. CA: SAGE Publications Ltd.
- Tedeschi, R. G., & Calhoun, L. G. (1996). The Posttraumatic Growth Inventory: measuring the positive legacy of trauma. *Journal of Traumatic Stress*, 9(3), 455-471.
- Tedeschi, R. G., & Calhoun, L. G. (2004). Posttraumatic Growth: Conceptual Foundations and Empirical Evidence. *Psychological Inquiry*, 15(1), 1-18. doi: 10.1207/s15327965pli1501_01
- Toren, P., & Horesh, N. (2007). Psychiatric morbidity in adolescents operated in childhood for congenital cyanotic heart disease. *Journal of Paediatrics and Child Health*, 43(10), 662-666. doi: 10.1111/j.1440-1754.2007.01183.x
- van der Linde, D., Konings, E. E., Slager, M. A., Witsenburg, M., Helbing, W. A., Takkenberg, J. J., & Roos-Hesselink, J. W. (2011). Birth prevalence of congenital heart disease worldwide: a systematic review and meta-analysis. *Journal of the American College of Cardiology*, 58(21), 2241-2247. doi: 10.1016/j.jacc.2011.08.025
- VERBI Software. (1989-2017). MAXQDA, software for qualitative data analysis (Version 12). Berlin, Germany.
- Vishnevsky, T., Cann, A., Calhoun, L. G., Tedeschi, R. G., & Demakis, G. J. (2010). Gender differences in self-reported posttraumatic growth: A meta-analysis. *Psychology of Women Quarterly*, 34(1), 110-120. doi: 10.1111/j.1471-6402.2009.01546.x
- Wang, Kai-Wei Katherine and Barnard, Alan (2004) Technology-dependent children and their families. *Journal of Advanced Nursing* 45(1):pp. 36-46

- Weathers, F. W., Litz, B. T., Keane, T. M., Palmieri, P. A., Marx, B. P., & Schnurr, P. P. (2013). The PTSD checklist for DSM-5 (PCL-5).
- Wei, H., Roscigno, C. I., Hanson, C. C., & Swanson, K. M. (2015). Families of children with congenital heart disease: A literature review. *Heart & Lung: The Journal of Acute and Critical Care*, 44(6), 494-511. doi: <https://doi.org/10.1016/j.hrtlng.2015.08.005>
- Wei, H., Roscigno, C. I., Swanson, K. M., Black, B. P., Hudson-Barr, D., & Hanson, C. C. (2016). Parents' experiences of having a child undergoing congenital heart surgery: An emotional rollercoaster from shocking to blessing. *Heart & Lung: The Journal of Acute and Critical Care*, 45(2), 154-160.
- World Health Organisation. (1992). *The ICD-10 classification of mental and behavioural disorders: Clinical descriptions and diagnostic guidelines* (10th Revision ed.). Geneva: World Health Organisation.
- Wortman, C. B. (2004). Posttraumatic Growth: Progress and Problems. *Psychological Inquiry*, 15(1), 81-90. doi: 10.2307/20447207
- Wray, J., Lee, K., Dearmun, N., & Franck, L. (2011). Parental anxiety and stress during children's hospitalisation: the StayClose study. *J Child Health Care*, 15(3), 163-174. doi: 10.1177/1367493511408632
- Wray, J., & Sensky, T. (2004). Psychological functioning in parents of children undergoing elective cardiac surgery. *Cardiology in the Young*, 14(2), 131-139. doi: 10.1017/s1047951104002045
- Wren, C., & O'Sullivan, J. J. (2001). Survival with congenital heart disease and need for follow up in adult life. *Heart*, 85(4), 438-443.

Appendix A: PTSD in parents of children with CHD literature search flow chart

Appendix B: Literature search strategy and terms for posttraumatic growth in parents of children with CHD

A systematic literature search of the electronic databases was conducted using the following parameters

Parameters	
Databases searched	PsycINFO, Psychology and Behavioral Sciences Collection, PsycARTICLES, MEDLINE, CINAHL with Full Text.
Time frame searched	from the earliest date for each database, until April 2016.
Search terms used	('congenital') AND ('heart defect' OR 'heart disease' OR 'cardiac' OR 'anomaly' OR, 'lesion' OR 'malformation') AND ('parent*' OR 'mother*' OR 'maternal' OR 'father*' OR 'paternal' OR 'caregiver') AND ('posttraumatic growth' OR 'posttraumatic growth' OR 'posttraumatic growth' OR 'posttraumatic change' OR 'posttraumatic change' OR 'posttraumatic change' OR "stress related growth" OR "adversarial growth" OR "positive psychological changes" OR ("thriving AND "trauma") OR ("benefit finding" OR "perceived benefits" AND 'trauma')
Limits applied	Articles published in English

Appendix C: Study Information Sheet for Website

What is the aim of this study?

This study is being conducted as part of a doctoral qualification in Clinical Psychology at the University of Essex -Tavistock training programme

The aim of this study is to better understand how parents of children with congenital heart conditions make sense of difficult and distressing experiences during their child's treatment. We would like to find out how this affects them and how they cope.

Before you decide whether or not to take part it is important for you to understand what this study is about and what taking part will mean for you. Please take time to read the following information carefully and discuss it with others if you wish. Ask us if there is anything that is not clear. For further information on this study please contact Emily Tan on the contact details below.

Why have I been invited to take part?

We are inviting you to take part in this study because we understand from the Children's Heart Federation that you might be interested in this topic. We are looking for parents of children with a diagnosis of a congenital heart condition.

How do I get involved?

This is a two part study. The first part is open to all parents of children with a diagnosis of a congenital heart condition without any other diagnosed developmental disorders. This part of the study involves answering four quick questionnaires accessible only through an online link.

The second part of the study involves participating in an online discussion group. We are looking to hear from parents with a range of different experiences. We will select parents from those who have completed the questionnaires and are interested in sharing their experiences to form small online discussion groups. If you are interested in taking part in this part of the study you will be asked to leave an email contact address through the online link. It is possible not everyone will be selected to participate in an online discussion group. The discussion groups will be conducted through closed Facebook forums. You do not have to participate in the online discussion group if you do not want to.

How does the link to the questionnaires work?

The link will take you to an online dedicated survey page. Your responses will be kept secure and will only be accessible to the researchers. Only the information you enter into the survey page will be accessible to the researchers.

How do the Facebook forums work?

The discussion forums are closed groups so only other members of the same forum group and the researchers will be able to identify you from your posts. Please note that no names will be given out to anyone. This type of group is known as a "secret"

group on Facebook. This means that only members of the group have access to the group and can see the group. The group will not be visible on your Facebook profile to other people on Facebook.

What will happen if I join the Facebook discussion forums?

If you would like to join the discussion forum on Facebook you will need to have a Facebook account. You will be asked to accept the Facebook administrator as a friend on Facebook. You will then be given access to the group and can begin posting in the discussion.

A new question is posed on the forum each week for 6 weeks and you will have a week to respond to the question. There is no right or wrong answers to these questions. We are interested in your perspective and thoughts on each question. You should also feel free to discuss the question with other people in the forum or comment on similarities or differences in perspective or experience.

The discussion forums will be moderated by Emily Tan and participants will be asked to adhere to the rules of the forum. Posts to the forum which break these rules may be removed and participants who repeatedly disregard the rules may be asked to leave the group.

After the discussion has been analysed the themes from the discussion will be posed back to the group for group members to comment on. After this the group will be closed. This means all the content of the group will be removed from Facebook by the group administrator. Then all members including the group administrator will be removed from the group. The group will no longer appear visible to you on Facebook. Facebook automatically deletes groups with no members.

Who is the study open to?

We would like to hear from parents over the age of 18, who are raising or have experienced raising a child diagnosed with a congenital heart condition.

What if I change my mind and I don't want to carry on with the study (I want to withdraw)?

If you choose to take part and then decide you would prefer to withdraw, then you can tell Emily Tan. You can leave the study at any time. You do not have to explain why and you do not have to give us any more information. This will not affect the support you receive from the Children's Heart Federation or any other services in any way.

If you do not wish us to use the information you have given us, we will destroy the information. If you wish to withdraw your questionnaire responses you can contact Emily Tan who will ensure that your data is removed from the study. You can also leave the Facebook discussion forum at any time. If you wish your posts to be removed from the forum and excluded from the study you can do this by contacting Emily Tan who will ensure that your posts are removed. You will also be removed from the group and will not have access to the group discussion posts.

How will the information be used?

The results of this study will be reported in a doctoral thesis. The findings will be used to develop better ways of providing parents with information about potentially distressing events and what they might feel. The findings may be presented through Children's Heart Federation website, in the media and in scientific reports and papers. We may use direct quotes in an anonymised form when reporting the results of the study in publications and presentations. A summary of the research findings will be sent to all the participants who wish to receive this at the end of the study.

Who has reviewed the study?

This study has been reviewed by The University of Essex Ethics Board (Ref. 14029)

What are the possible benefits of taking part?

There may not be any direct benefit to you of taking part but we hope that the results from this study will be used to develop better ways of providing parents with information about distressing events and what other parents have experienced. Some parents may find discussing their experiences with others in a similar situation helpful; however this is not the primary aim of the discussion group.

What are the possible disadvantages and risks of taking part?

There are no real risks or disadvantages to taking part. We should note that it is possible that important information relevant to this topic could come from bereaved parents. As such, some discussions may touch upon sensitive issues for some families. If you should feel uncomfortable talking about or reading other parent's experience of distressing events related to their child, or do not want to continue taking part you are free to stop at any time.

If you are affected by any of the discussions on the forum and would like to talk to someone you can contact the Children's Heart Federation Helpline 0808 808 5000 or email info@chfed.org.uk who will also be able to direct you to further sources of support if you wish.

What if there is a problem?

If in the event that there is evidence to suggest that you might be experiencing levels of distress which might pose a risk to your future wellbeing you will be contacted by the researcher by email and advised to seek professional medical consultation.

If you have concerns about the study, then you can speak or write to Emily Tan or her academic supervisors, details below.

Will my General Practitioner (GP) know I am taking part?

We do not propose to inform your GP about your participation.

Will my taking part in the study be kept confidential?

Your identity and participation in the study will be kept confidential. If you accidentally post personally identifying information to the Facebook discussion forum the moderator will remove the identifying information to protect your identity.

How will you store my data?

Your information will be anonymised and kept secure in an encrypted electronic format.

Further information and contact details

The study team are based at Essex University. If you would like to have further information about the study or wish to write or speak to someone about it, then please contact us:

Emily Tan, Trainee Clinical Psychologist

Email: ectan@essex.ac.uk

Dr Leanne Andrews, Academic supervisor

Email: landre@essex.ac.uk

Dr Fran Davies, Academic supervisor

Email: fdavies@essex.ac.uk

The School of Health and Human Sciences

University of Essex

Wivenhoe Park

Colchester, CO4 3SQ

Telephone: +44(0)1206 873910

What to do now

Please read through the information and talk about the study with family members, friends or colleagues if you wish. It is your choice whether to take part and you may contact Emily Tan on 0793 489 8789 or email ectan@essex.ac.uk if you need more information or would like to ask questions or talk to someone about the study before deciding.

If you have decided you want to take part...

If you wish to take part, then please click on the link to the questionnaires below

[Link to Questionnaires](#)

Thank you for taking the time to read this information

Appendix D: Facebook study advertisement wording

Exploring experiences, distress and coping of parents of children with congenital heart disease

The aim of this study is to better understand how parents of children with congenital heart conditions make sense of difficult and distressing experiences related to their child's condition and treatment. We would like to find out how this affects parent and how they cope. It is hoped this will help us find better ways of providing parents with information about what they might feel and how they might be supported to cope better.

We would really like to hear from parents. There will be two parts to the study: questionnaires and small closed Facebook discussion groups. If you would like to participate you can click on the link to the Children's Heart Federation website below for further information.

LINK TO STUDY WEBPAGE INFORMATION SHEET

If you have any questions you can contact Emily Tan on 0793 489 8789 or by email ectan@essex.ac.uk

This study is being conducted as part of a doctoral qualification in Clinical Psychology at the University of Essex -Tavistock training programme.

Appendix E: Demographic Questions

Question	Response key
Your Gender	Male Female Other Do not wish to disclose
Your Age	
Your Ethnicity	White British White Other Mixed Black or Black British Asian or Asian British Any other Do not wish to disclose
Your Child's Age	
Your Child's Congenital Heart Problem(s)	
When was your child's last treatment	Less than a month ago Between 1 – 3 months ago Between 3 – 6 months ago Between 6 – 12 months ago More than 1 year ago More than 2 years ago More than 5 years ago More than 10 years ago
Please list any other developmental disorders or physical disabilities your child has eg Learning Disability	
Please describe your family home structure. Eg Your child lives with: one parent, two parents, spends time living with each parent, lives with other family members.	
Number of other children in the family	

Appendix F: The Perceived Ability to Cope with Trauma (PACT) scale

Sometimes we must contend with difficult and upsetting events. Unfortunately, sometimes we are confronted with events that might be traumatic and disruptive to the course of our lives. Examples of such events include the death or injury of someone close to us, a natural disaster, a serious accident or illness, sexual and physical assault, and terrorist attack. Below you will find a list of different kinds of behaviours and strategies that people sometimes use in the weeks following potentially traumatic events. This questionnaire asks which of these behaviours and strategies you might be able to use. Please rate the extent that you **would be able to use** each of these behaviours and strategies following a potentially traumatic event if you needed to.

	Focus *	Behaviours and Strategies I would be able to.....	Not True Extremel y True						
1	T	Let myself fully experience some of the painful emotions linked with the event	1	2	3	4	5	6	7
2	F	Keep my schedule and activities as constant as possible	1	2	3	4	5	6	7
3	F	Comfort other people	1	2	3	4	5	6	7
4	T	Pay attention to the distressing feelings that result from the event	1	2	3	4	5	6	7
5	F	Remind myself that things will get better	1	2	3	4	5	6	7
6	F	Distract myself to keep from thinking about the event	1	2	3	4	5	6	7
7	T	Reflect on the meaning of the event	1	2	3	4	5	6	7
8	F	Look for a silver lining	1	2	3	4	5	6	7
9	F	Find activities to help me keep the event off my mind	1	2	3	4	5	6	7
10	T	Remember the details of the event	1	2	3	4	5	6	7
11	F	Keep myself serious and calm	1	2	3	4	5	6	7
12	T	Alter my daily routine	1	2	3	4	5	6	7
13	F	I would be able to laugh	1	2	3	4	5	6	7
14	F	Focus my attention on or care for the needs of other people	1	2	3	4	5	6	7
15	F	Enjoy something that I would normally find funny or amusing	1	2	3	4	5	6	7
16	T	Spend time alone	1	2	3	4	5	6	7
17	F	Try to lessen the experience of painful emotions	1	2	3	4	5	6	7
18	T	Reduce my normal social obligations	1	2	3	4	5	6	7
19	T	Face the grim reality head on	1	2	3	4	5	6	7
20	F	Stay focused on my current goals and plans	1	2	3	4	5	6	7

*T = Trauma focussed coping, F = Forward focussed coping

Taken from Bonanno, Pat-Horenczyk and Noll (2011) with permission.

Appendix G: The PTSD Checklist for DSM-5 PCL-5

Instructions: Below is a list of problems that people sometimes have in response to a very stressful experience. Please read each problem carefully and then circle one of the numbers to the right to indicate how much you have been bothered by that problem in the past month.

	<i>In the past month how much were you bothered by</i>	Not at all	A little bit	Moderately	Quite a bit	Extremely
1	Repeated, disturbing, and unwanted memories of the stressful experience?	0	1	2	3	4
2	Repeated, disturbing dreams of the stressful experience?	0	1	2	3	4
3	Suddenly feeling or acting as if the stressful experience were actually happening again (<i>as if you were actually back there reliving it</i>)?	0	1	2	3	4
4	Feeling very upset when something reminded you of the stressful experience?	0	1	2	3	4
5	Having strong physical reactions when something reminded you of the stressful experience (<i>for example, heart pounding, trouble breathing, sweating</i>)?	0	1	2	3	4
6	Avoiding memories, thoughts, or feelings related to the stressful experience?	0	1	2	3	4
7	Avoiding external reminders of the stressful experience (<i>for example, people, places, conversations, activities, objects, or situations</i>)?	0	1	2	3	4
8	Trouble remembering important parts of the stressful experience?	0	1	2	3	4
9	Having strong negative beliefs about yourself, other people, or the world (<i>for example, having thoughts such as: I am bad, there is something seriously wrong with me, no one can be trusted, the world is completely dangerous</i>)?	0	1	2	3	4
10	Blaming yourself or someone else for the stressful experience or what happened after it?	0	1	2	3	4
11	Having strong negative feelings such as fear, horror, anger, guilt, or shame?	0	1	2	3	4
12	Loss of interest in activities that you used to enjoy?	0	1	2	3	4
13	Feeling distant or cut off from other people?	0	1	2	3	4
14	Trouble experiencing positive feelings (<i>for example, being unable to feel happiness or have loving feelings for people close to you</i>)?	0	1	2	3	4
15	Irritable behavior, angry outbursts, or acting aggressively?	0	1	2	3	4
16	Taking too many risks or doing things that could cause you harm?	0	1	2	3	4
17	Being “superalert” or watchful or on guard?	0	1	2	3	4
18	Feeling jumpy or easily startled?	0	1	2	3	4
19	Having difficulty concentrating?	0	1	2	3	4
20	Trouble falling or staying asleep?	0	1	2	3	4

PCL-5 (8/14/2013) Weathers, Litz, Keane, Palmieri, Marx, & Schnurr --National Center for PTSD

**Appendix H: The Psychological Well-Being – Posttraumatic changes questionnaire
(PWB-PTCQ)**

Listed below are 18 statements. Please mark the appropriate box beside each description indicating how much you feel you have changed as a result of your child's heart condition. The 1 to 5 scale is as follows:

- 1** = Much less so now
2 = A bit less so now
3 = I feel the same about this as before
4 = A bit more so now
5 = Much more so now

		1	2	3	4	5
1	I like myself.					
2	I have confidence in my opinions.					
3	I have a sense of purpose in life.					
4	I have strong and close relationships in my life.					
5	I feel I am in control of my life.					
6	I am open to new experiences that challenge me.					
7	I accept who I am, with both my strengths and limitations.					
8	I don't worry what other people think of me.					
9	My life has meaning.					
10	I am a compassionate and giving person.					
11	I handle my responsibilities in life well.					
12	I am always seeking to learn about myself.					
13	I respect myself.					
14	I know what is important to me and will stand my ground, even if others disagree					
15	I feel that my life is worthwhile and that I play a valuable role in things					
16	I am grateful to have people in my life who care for me.					
17	I am able to cope with what life throws at me. .					
18	I am hopeful about my future and look forward to possibilities.					

(Joseph et al., 2012)

Appendix I: Consent form for participants

(First page of electronic survey)

Please complete this form after you have read the information about the study on the Children's Heart Federation Website (*LINK TO STUDY INFORMATION PAGE*).

**Title of Study "Exploring traumatic experiences, posttraumatic growth and coping flexibility in parents of children with congenital heart disease:
A mixed methods study"**

University of Essex Research Ethics Approval Number:

Thank you for considering taking part in this research. If you have any questions arising from the information sheet, please contact Emily Tan before you decide whether to take part. You are advised to keep a copy of the information sheet for your records.

- | | Please check the
box |
|---|---------------------------------|
| 1. I confirm that I have read and understand the information pages for the above study and have had the opportunity to ask questions. | <input type="checkbox"/> |
| 2. I understand that my participation is voluntary and that I am free to withdraw at any time, without giving reason and without my legal rights being affected | <input type="checkbox"/> |
| 3. I understand that if I decide at any time before or during the research that I no longer wish to participate in this project, I can notify the researchers involved and withdraw from it immediately without giving any reasons. I can request for information I have contributed to be excluded from the study. | <input type="checkbox"/> |
| 5. I understand that my personal information will be treated as strictly confidential and will be handled in accordance with the terms of the Data Protection Act 1998. | <input type="checkbox"/> |
| 6. I consent to completing questionnaires. | <input type="checkbox"/> |
| 7. I consent to sharing my experiences in a closed group and reading other parent's experiences of events related to their child which may be distressing. | <input type="checkbox"/> |
| Please enter you email address below | |
| 8. I understand that I may not be selected to participate in a closed discussion group. | <input type="checkbox"/> |

For further details contact: Emily Tan, Trainee Clinical Psychologist, Principal Investigator, ectan@essex.ac.uk School of Health and Human Sciences, University of Essex, Wivenhoe Park, Colchester, CO4 3SQ

Appendix J: Discussion group joining email wording

Dear Parent,

Thank you for participating in the emotional coping strategies study and completing the online questionnaires. I am writing to you to invite you to join an online closed group discussion. The discussions will be held on Facebook using “secret” groups. This means that only members of the group have access to the group and can see the group. The group will not be visible on your Facebook profile to other people on Facebook.

A new question is posed on the forum each week for 6 weeks and you will have a week to respond to the question. There is no right or wrong answers to these questions. We are interested in your perspective and thoughts on each question. You should also feel free to discuss the question with other people in the forum or comment on similarities or differences in perspective or experience.

The discussion forums will be moderated by Emily Tan and participants will be asked to adhere to the rules of the forum. Posts to the forum which break these rules may be removed and participants who repeatedly disregard the rules may be asked to leave the group.

After the discussion has been analysed the themes from the discussion will be posed back to the group for group members to comment on. After this the group will be closed. This means all the content of the group will be removed from Facebook by the group administrator. Then all members including the group administrator will be removed from the group. The group will no longer appear visible to you on Facebook. Facebook automatically deletes groups with no members.

If you would like to participate:

1. You will need to have a Facebook account.
2. Find “Emily Tan Emotional Coping Strategies Study” on Facebook and add me as a friend
3. I will then add you to a small closed discussion group. You will then be given access to the group and can begin posting in the discussion.

Please find attached some guidelines for using the online closed group.

Please find a link to the study information page below
<http://www.chfed.org.uk/emotional-coping-strategies-study/>

If you have any further questions please do not hesitate to contact me on 0793 489 8789 or by email
ectan@essex.ac.uk

Looking forward to hearing from you.

Appendix K: Discussion group guidelines

Keeping the forum secure

If you are using a computer or device which others also use please remember that others may be able to access your Facebook page too.

Please remember to log out of Facebook before closing your internet browser or App. (You may need to untick the *keep me logged in* option on the login page for the duration of the study)

Answering questions

There is no right or wrong answers to the weekly questions. We are interested in your experiences, perspectives and thoughts on each question.

You should also feel free to discuss the question with other people in the forum or comment on similarities or differences in perspective or experience.

If you accidentally post identifying information the moderator will remove your post and repost it replacing it with XXX.

Group rules

- Be respectful of other people in the group – no abusive language
- Do not post identifying personal information such as your phone number or address
- Do not post the names of health professionals or hospitals involved in your child's care
- If you have any concerns immediately contact the moderator

If you have any questions you can contact Emily Tan on 0793 489 8789 or by email ectan@essex.ac.uk

Appendix L: Example of coding frame

MAXQDA12		
Code System		
<input type="checkbox"/>	Child death - grief	1
<input type="checkbox"/>	Facing possibility of Child's death	30
<input type="checkbox"/>	Child distress	22
<input type="checkbox"/>	Child Anxiety	1
<input type="checkbox"/>	Child - young/small	2
<input type="checkbox"/>	Child - coping well	1
<input type="checkbox"/>	Child longer term impact	12
<input type="checkbox"/>	Healthy child	5
<input type="checkbox"/>	Previously healthy	2
<input type="checkbox"/>	Parents - expectations	3
<input type="checkbox"/>	Parents - health	4
▶ <input type="checkbox"/>	Parental role - provider	6
<input type="checkbox"/>	Parent role - strong	7
<input type="checkbox"/>	Parent role - guide child	2
<input type="checkbox"/>	parent role - procedures	7
<input type="checkbox"/>	Parental role - protect	17
<input type="checkbox"/>	Parent role - info	5
<input type="checkbox"/>	Parental role - split	4
<input type="checkbox"/>	Parental - helplessness	9
<input type="checkbox"/>	Parent -Long lasting impact	6
<input type="checkbox"/>	delayed response	4
<input type="checkbox"/>	time to process	7
<input type="checkbox"/>	Physiological response	4
▶ <input type="checkbox"/>	Anxiety	29
<input type="checkbox"/>	Distress	27
<input type="checkbox"/>	Traumatic	28
<input type="checkbox"/>	Unexpected	21
<input type="checkbox"/>	Fear for the future	13
<input type="checkbox"/>	Uncertainty in the future	17
<input type="checkbox"/>	Not controllable	18
<input type="checkbox"/>	Coping - Getting on with it	9
<input type="checkbox"/>	Not knowing what happened/ was happening	28
▲ <input type="checkbox"/>	Knowing something was wrong	8
<input type="checkbox"/>	not realising something was wrong	1
<input type="checkbox"/>	Cyclical nature	25
<input type="checkbox"/>	Minimisation	25
<input type="checkbox"/>	Medical factual	30
▶ <input type="checkbox"/>	being prepared	6
<input type="checkbox"/>	Psychology/ counselling	9
<input type="checkbox"/>	Asking for help	6
<input type="checkbox"/>	Help/Support	17
▶ <input type="checkbox"/>	Recovery time	34
<input type="checkbox"/>	poor outcome	14
<input type="checkbox"/>	Good outcome	16
<input type="checkbox"/>	Defying Expectations +	5
<input type="checkbox"/>	Not allowing condition negative impact on life	2
<input type="checkbox"/>	BLUE	6

Appendix M: Example of reflective memos made during analysis

The screenshot displays the MAXQDA software interface. On the left, the 'Code System' pane shows a hierarchical list of codes, with 'Parental role - protect' selected. The 'Document System' pane on the top left shows a list of documents, with 'Group 1 LOW' selected. The 'Document Browser' pane on the top right shows a list of documents with their corresponding codes and line numbers. A 'Memo' window is open in the foreground, displaying the following text:

Memo

Code: Parental role - protect Linked codes

Title: Parental role - protect

Author: Emily Creation date: 29/02/2016 14:22

Type: [Color-coded icons]

Type label:

Calibri 11 B / U [Color-coded icons]

This also links to - being strong
isolating from other/ being removed from ed life

defending, standing up for, safeguarding - preventing - knowing st was wrong
taking on worries for others ie child/ partner

Protecting child but also others ie partner

The background shows a list of documents with their corresponding codes and line numbers. The 'Document Browser' pane shows a list of documents with their corresponding codes and line numbers. The 'Document Browser' pane shows a list of documents with their corresponding codes and line numbers.

49 **Moderator:** I just want to say thank you for all your time and contributions so far I realise that life can get really busy and it can be hard to find time, so thanks again. This week I'd like to ask you about how you got through those distressing situations and other difficult times. What helped you to get through and also what was not helpful? What would you have liked to be done differently by others? What would you have done differently yourself?

50 **Megan:** I had really good care even though the hdu staff and picu staff were there for XXX...they knew i was struggling physically(he was c section just 6 days before) and emotionally after loss of his brother year before. I never had to repeat anything they kept us all involved and even knew and talked to me about his sisters at home. They made sure i ate and on occasion forced me by hand out the door to rest lol. Having that personal touch to care really helped remove the fear of such a clinical setting i had and helped me not panic as much. I dont think anyone could have done anything differently, apart from maybe a little bit more help with a place to sleep. Straight after section a chair wasnt ideal for few days but i know ronald mcdonald beds were tight so couldnt complain really. I would have spoken to more people more families on the wards. With kids at home hubby was there i was alone with a very poorly 6 day old and although staff were ppa. Loneliness fuelled the fear that we were going to lose XXX too.

process that i couldnt describe how i did it...i didnt cope...i just learn to deal with it

side, I'm not sure how I would have coped on my own, I would have, but having helpd alot. It was one part I didn't have to stress about. Im sure many things happened weren't very helpful at the time, but the situation was difficult for all. I'm not sure what I roughly at the time by his doctors. I wish I had spoken out more when unhappy with g on the sidelines. I may come back to this one as I remember things smile emoticon

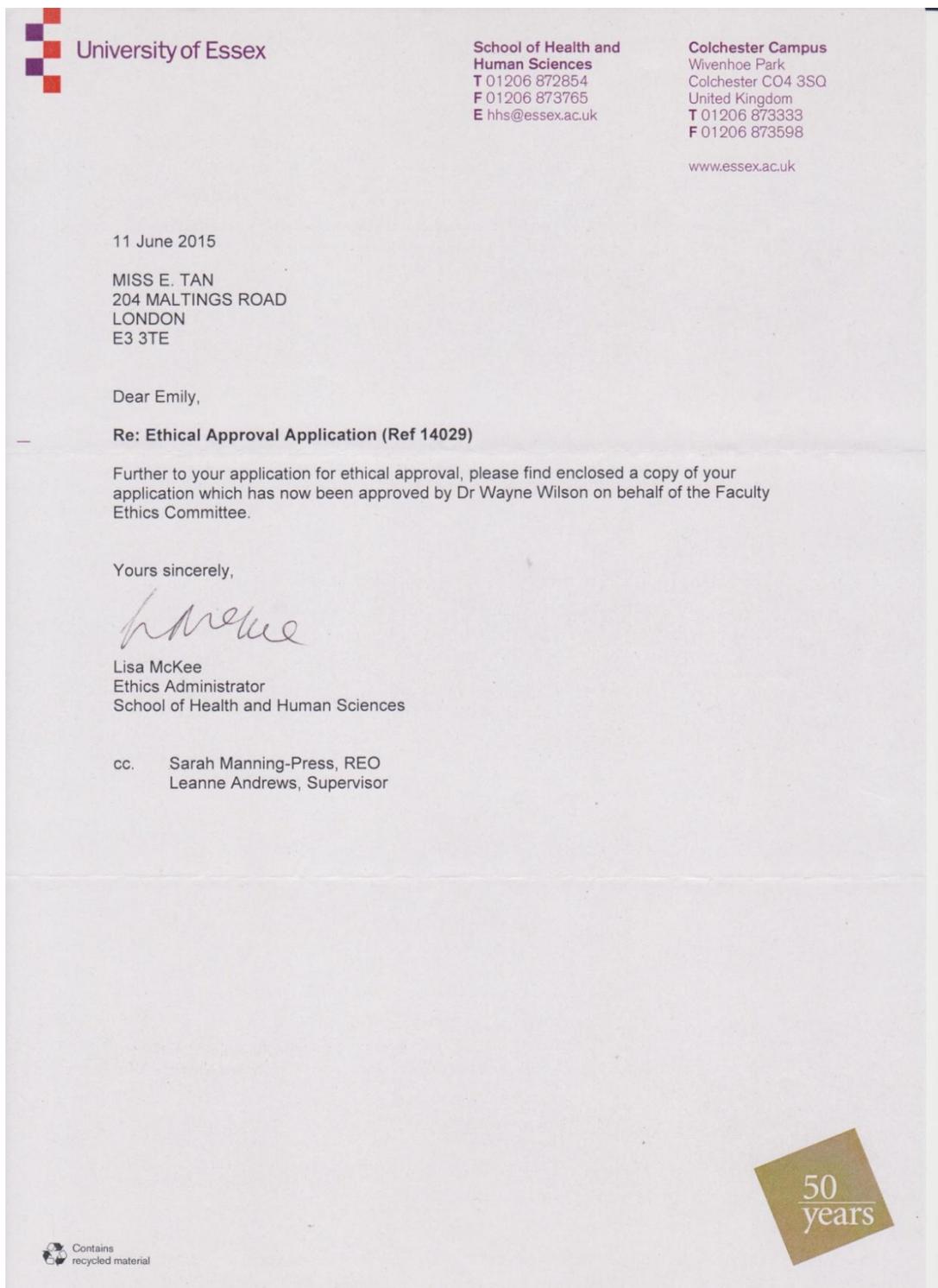
look after My daughters and my mother in law did all my washing and ironing so that bad when I left XXX. The nurses on ITU were brilliant, well everyone was, but they used to let us bath him which was brill seeing we couldn't hold him. They also kept us different was getting diagnosed from my scans (and I had my last one at 36weeks as he so I know how it works, we sort of pulled a few strings to get all his ve done anything differently as its all out of your control and felt like everything was very

already. This week I'd like to ask you about how you got through those distressing distance my self from friends and family. And wish there was more support from

professionals like counselling.

56 **Laura:** I struggled with the comments of others who simply did not get how serious it was. I was told several times 'oh my child's got asthma and the doctors always make it sound worse, they're fine really' - I felt like it demeaned my experience or that my reaction was over the top. I gave up explaining 'no, he nearly died'. I had one GP who was fab. She understood, gave me the time and took me seriously when I saw her about myself and my son in the weeks following. I also accessed counselling through work and this helped transition back to work. Medication also really helped. My health visitor was useless, never acknowledged that she ignored my concerns in the first place. Our relationship deteriorated and I eventually told them they weren't welcome in my house. When my mat leave ended I was petrified of going back. I was working full time and we couldn't afford for me to take another months mat leave or to reduce my working hours. In reality going back was the best thing for me. With a few weeks I felt hugely better with something else to focus on other than my son, what he'd been through and what the future held. During the bad times I kept some things from my husband - he didn't entirely see the seriousness of some things (as a nurse I saw things differently) and I decided to protect him. I know now that made it worse for me. I felt I was carrying everything myself and had to hold it together for the sake of my family. A couple of years ago my father in law was seriously ill - I made the conscious choice then that I would be completely honest with my husband about my opinion on the situation. It was easier to carry that bad experience together (FIL died a few days later)

57 I know I said before but I feel being a nurse impacted on my experience. I know whilst it was going on I was getting messages from non nurse friends saying "it must be so much better for you being a nurse and understanding everything that is going on". At the same time nurse friends were saying "it must be so much worse as you know fully what is going on" - I definitely identified

Appendix N: Ethical Approval

Appendix O: Parent reported child heart problems

Septal Defects

- Atrial Septal Defect
- Atrioventricular Septal Defect
- Ventricular Septal Defect
- Patent ductus arteriosus

Defects of the outflow tracts and great vessels

- Tetralogy of Fallot
- Pulmonary Atresia
 - with Ventricular Septal Defect
 - with intact Ventricular Septum
- Transposition of the Great Arteries
- Truncus Arteriosus
- Pulmonary Stenosis
- Coarctation of the Aorta/ Critical Aortic Stenosis /Aortic Stenosis
- Subaortic stenosis
- Ebstein's Anomaly
- ALCAPA Anomalous left coronary artery arising from the pulmonary artery
- Interrupted Aortic arch/ aortic arch syndrome
- Aortopulmonary window

Univentricular Hearts

- Hypoplastic right heart
- Hypoplastic left heart syndrome
- Double inlet left ventricle
- Double inlet right ventricle

Valves

- Mitral valve leaks/ prolapse
- Pulmonary valve leaks/ regurgitation
- Partial pulmonary venous drainage
- Tricuspid atresia
- Bicuspid valve

Other heart problems

- Dextrocardia & Right atrial isomerism
- Cardiomyopathy/ Dilated cardiomyopathy

Other heart symptoms

- Left bundle branch block
- Pulmonary hypertension