UNIVERSITY OF SOUTHERN QUEENSLAND

THE EXPERIENCE OF PSYCHOLOGICAL DISTRESS, SYMPTOMS OF TRAUMA, COPING AND POST-TRAUMATIC GROWTH IN PARENTS OF CHILDREN WITH CONGENITAL HEART DISEASE: A SYSTEMATIC REVIEW OF LITERATURE

A thesis submitted by

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For the award of

Master of Clinical Psychology

2017

Abstract

AIM: This systematic review explored the experience of psychological distress, trauma, post-traumatic growth (PTG) and coping among parents of children with congenital heart disease (PCCHD).

METHOD: PRSIMA guidelines for systematic reviews were followed to identify 82 studies for inclusion and analysis.

FINDINGS: PCCHD experience high levels of distress that manifest in a variety of ways (including depression, anxiety, stress and somatization). Generally, the distress occurs at levels higher than normative populations and at levels consistent with parents of children with other chronic and/or severe health conditions. Psychosocial stressors (commonly but not exclusively found in families of more severe CHD) appear to exacerbate this experience. Diagnosis, and birth, of a child with CHD is deeply distressing for PCCHD. Invasive surgical procedures (e.g. open-heart surgery) are also a significant source of distress for PCCHD, regardless of CHD severity or the complexity of the procedure, and may serve as a trigger for concerns about child mortality and suffering, and the experiences of role loss, uncertainty and lack of control. Parental descriptions of these experiences are consistent with those of other survivors of traumatic experiences. A number of PCCHD meet threshold for diagnosis of Acute Stress Disorder and Post-Traumatic Stress Disorder and many more experience multiple clinically significant symptoms. Ongoing symptoms of trauma (in particular, hypervigilance), depression and anxiety are also frequently reported. PCCHD employ a variety of coping strategies in an attempt to manage the effects of their exposure to CHD-related trauma. Concurrently, they also appear to experience the transformational nature of this trauma in a way that is consistent with the features of PTG.

DISCUSSION: This review confirms and extends the existing literature regarding the psychological experiences of families of children with CHD. In addition, it highlights the

traumatic nature of these events and identifies that symptom trajectories may vary as a function of time and type. This review also uniquely identifies evidence suggesting that PCCHD experience not just the distressing symptoms of trauma exposure, but also the transformational features suggestive of PTG. Findings are discussed in relation to existing research and implications for further research and practice.

Certification of Dissertation

This thesis is entirely the work of Jodie Brown, except where otherwise acknowledged. The work is original and has not previously been submitted for any other award, except where acknowledged.

Student and supervisors' signatures of endorsement are held at USQ.

Acknowledgements

A thesis is rarely completed without the assistance and support of a multitude of people and such was the case with this one. As such, I would like to acknowledge the many academic, administrative and support staff of the University who supported me throughout my postgraduate studies and who inspired me with their knowledge, enthusiasm and care. A sincere thank you to my supervisor Dr Amy Mullens for her guidance throughout the preparation of this thesis, for her comments on drafts of this manuscript and for her support and encouragement throughout the entire process. Thank you, also, to Dr Erich Fein for his advice on the final preparation of the manuscript.

A special thank you to my family and friends for their support and belief in me (and for not rolling their eyes too often when I'd mention "my thesis" yet again!). In particular, to my children, Jack and Lily, for reminding me of what is important and for their love and patience as 'Mummy' juggled her many roles and responsibilities; to my partner Michael for being my rock, always believing in me and living the definition of 'partnership' every day; to my father and mother-in-law for providing hours of babysitting; to Pradhayini for unswerving support, encouragement and friendship; to my cousin and friend Kim for being my constant 'cheer-squad' despite her own challenges and busy family life; and to the 2015 Clinical Masters cohort for sharing this journey with me.

Finally, I would like to thank all of the families who took part in the research reviewed for this thesis – because of your bravery in sharing your experiences, we can work together to better understand the 'heart journey' and improve the support available for CHD children and their families.

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Chapter 1- Introduction

1.1 Context and Background

Three quarters of Australian infant deaths are attributable to perinatal complications and congenital defects, with Congenital Heart Disease (CHD) responsible for twenty percent of perinatal deaths (AIHW, 2017; Al-Yaman, Bryant & Sargeant, 2002). CHD refers to any malformation (structural or functional) of the heart, valves and/or central blood vessels that is present at birth (CDC, 2017; Hoffman, 1995; Leggat, 2011; NIH, 2011). As such, CHD comprises multiple disorders that vary greatly in severity and medical management, as well as prevalence (CDC, 2016 & 2017; Dodge-Khatami, Mavroudis & Backer, 2014; Hoffman & Kaplan, 2002; Leggat, 2011; NIH, 2011; Triedman & Newberger, 2016). CHDs can be classified by the structural complexity of the defect (Hoffman & Kaplan, 2002; Leggart, 2011; NIH, 2011;). Less complex defects (like a small Ventricular Septal Defect, VSD) may resolve without intervention. Moderate defects (such as a large Atrial Septal Defect, ASD) may require 'repair' through open-heart surgery and/or cardiac catheterisation. Complex defects (such as Hypoplastic Left Heart Syndrome, HLHS) are those more likely to be palliative in nature and to require lifelong care, multiple open-heart surgeries/cardiac catheterisations, and/or heart transplant. CHDs can also be classified based upon the presence or absence of cyanosis (reduced blood oxygen saturation, usually clinically evident at <85%), with cyanotic patients amongst those with increased complexity and medical vulnerability (Heinzman, 2009; Hoffman & Kaplan, 2002).

Types of CHD differ in frequency. Simple acyanotic defects such as VSDs and ASDs are the most common, comprising between 20-34% and 13-20% of CHDs, respectively (NHS, 2015; van der Linde et al., 2011). More complex cyanotic CHDs tend to be rarer, with conditions such as Truncus Arteriosus (TA) and Interrupted Aortic Arch (IAA) comprising less than one percent of CHDs (CDC, 2017; NHS, 2015). In a meta-analysis conducted in

2011, van der Linde et al. found that the eight most common CHDs were: VSD (34%); ASD (13%); Patent Ductus Arteriosus (PDA, 10%); Pulmonary Stenosis (PS, or Pulmonary Valve Stenosis, PVS, 8%); Coarction of the Aorta (CoA or COARC, 5%); Tetralogy of Fallot (TOF, 5%); Transposition of the Greater Arteries (TGA, 5%); and Aortic Stenosis (AoS, or Aortic Valve Stenosis, AVS, 4%).

Whilst many heart defects, simple and complex, require open heart surgery during childhood, approximately 25% of heart defects are considered to be critical in nature, with the patient requiring surgical intervention within the first hours to the first twelve months of life (Oster et al., 2013). Fifty percent of infants and children with CHD will require at least one cardiac surgery during their childhood (Heartkids, 2017; Hoffman & Kaplan, 2002). Table 1.1 provides details of CHD types, their relative percentage in relation to CHDs overall, prevalence rates, cyanotic status and complexity (Tables A1 and A2 in Appendix A include additional definitions/abbreviations).

1.1.1 Rates of CHD

Worldwide, the overall birth-rate for CHD is currently stable at approximately one in a hundred live births (Hoffman, 2013; Hoffman & Kaplan, 2002; Reller et al., 2008). Whilst overall variations in rate and type of CHD between and within nations were found to be non-significant in a 2011 meta-analysis, some studies have continued to report variations (Bjornard et al., 2013; CDC, 2016; British Heart Foundation, 2013; Hoffman, 2013; van der Linde et al., 2011). Within Australia, 3000 infants are born with CHD per annum (or approximately eight per day) and 32,000 children live with CHD (Heartkids, 2017; Hoffman & Kaplan, 2002). Whilst birth-rates may remain stable, it is anticipated that the number of children living with CHD may rise based upon an increasing number of births per year (AIHW, 2008; Tough et al., 2002); increasing survival rates for children with CHD (Leggat, 2011); increases in maternal age at time of pregnancy, with associated increases in risk of

congenital defects in the foetus (Reefhuis & Honein, 2004; Tough et al., 2002); and improved methods for early diagnosis (AIHW, 2008; Leggat 2011).

1.1.2 CHD mortality rates

Whilst CHD birth-rates are stable, mortality rates in developed countries have shown a significant decline over time. Between 1979-2008 in the United Kingdom, there was a decline in CHD-related deaths of 83%, with the downwards trend continuing from 2008-2013 (British Heart Foundation, 2013). In 2011, CHD accounted for 4.7% of all deaths of girls under the age of one and 3.5% of those in boys (British Heart Foundation, 2013). However, despite the greatest decline in deaths occurring in those under twelve months, of those that died of CHD in 2011, 47% were under the age of one (British Heart Foundation, 2013). In Australia, the largest number of deaths in infants under one remains attributable to CHD (Leggat, 2011; Heartkids, 2017).

Declines in mortality rates are likely due to more sophisticated diagnostic and surgical procedures. For example, within the United Kingdom, there was a 60% increase in surgeries performed for CHD between 2000 and 2010 (British Heart Foundation, 2013). Within the US, 25% of individuals born with CHD will have defects critical enough to require life-saving surgery before the age of one (Gilboa et al., 2016; Hoffman & Kaplan, 2002; Reller et al., 2008). Hospitalisations for CHD comprise 14.6% of all hospitalisations for congenital malformations, with CHD responsible for nearly 1% of all hospitalisations (AIHW, 2008; Kohler et al., 2005).

Table 1.1

CHD Types, Classification and Frequency Rates

Type of CHD	Description	% of CHD	Classification
Atrial Septal Defect (ASD) [1,5, 6,7,8, 14]	One or more holes between the two atria (upper chambers of the heart). Treatment depends on the size and location of the hole/s. Small holes often self-correct over time. Other ASDs require cardiac catherization or surgery. Timing depends on severity.	13-20	A S
Aortic Stenosis (AoS) or Aortic Valve Stenosis (AVS) [1, 6,7,8, 13]	Narrowing of the aortic valve with restricted blood flow and increased pressure in the left atrium	4-5	A S
Coarction of the Aorta (Coarc or CoA) [1, 6, 7, 8, 12]	Narrowing of part of the aorta that blocks normal blood flow to the body, causing thickening (and then weakening) of the heart muscle and, often, high blood pressure in the upper body and low blood pressure in the lower body. Intervention upon diagnosis - surgery or balloon angioplasty. Possible follow up surgeries and medication required.	5-12	A C*
Complete Atrioventricular Canal Defect (CAVC) or Endocardial Cushion Defect or Atrioventricular Septal Defect (AVSD) [3.5.14]	Malformation of heart valves and holes between the right and left chambers of the heart. May be complete (large hole in the center of the heart causing blood to mix between all four chambers) or partial (hole near the centre of the heart with associated damage to one of more valves). One or more surgeries - timing depends on severity. Can be cyanotic (complete) or acyanotic (some partial)	3-4.76	A/Cy C*
Double Outlet Right Ventricle (DORV) [4, 17, 18]	Both aorta and pulmonary artery connect to the right ventricle with an associated VSD that allows blood to reach the great arteries. Associated with increased blood flow and high pressure in the lung circulation system	1-3	Cy C*
Ebstein's Anomaly [6,7, 19]	Abnormal triscupid valve and right ventricle.	<1	Cy C*
Interrupted Aortic Arch (IAA) [12]	No connection between the two parts of the aortic arch, leading to reduced blood flow to the body (similar to CoA).	<1	Cy C*
Patent Ductus Arteriosus (PDA) [1, 6,7,8, 13]	Failure of the patent ductus arteriosus (a hole in the aorta that allows foetus' blood to bypass the lungs prior to birth) to close after birth, increasing pressure on the heart and lungs. Intervention is based upon size and severity with interventions ranging from monitoring and/or medication to surgery and/or catheterisation.	<1-10	A S
Pulmonary Stenosis (PS) or Pulmonary Valve Stenosis (PVS) [1, 6,7,8]	Narrowing of the pulmonary valve with high pressure in the right ventricle	8-10	A S

Single ventricle defects			
Hypoplastic Left Heart Syndrome (HLHS)[2,5, 6,7,8 , 14]	Left side of the heart is not fully developed. More specifically: underdeveloped and small left ventricle, mitral valve, aortic valve and ascending portion of the aorta, with (often) ASD. Multiple surgical interventions beginning soon after birth, medications and lifelong care.	2-9	Cy C*
Tricuspid Atresia [6,7]	Missing tricuspid valve with resultant under-developed right ventricle.	<1	Cy C*
Pulmonary Atresia/Intact Ventricular Septum [5, 6, 14]	Pulmonary valve (controls blood flow from the right ventricle to the pulmonary artery) is not formed. Can also include a VSD. Medication is required in the newborn to keep the ductus arteriosus open and catheterisation may be used to increase blood flow. In most cases surgical repair of the valve soon after birth is required. In severe cases with underdeveloped right ventricles, staged surgeries may also be required (similar to those for HLHS)	<1	C*
Tetralogy of Fallot (TOF) [1, 5, 6, 7, 8, 14]	Four defects: ventricular hypertrophy (thicker wall of lower right chamber); pulmonary stenosis (narrowing of pulmonary valve and main pulmonary artery); VSD (hole between lower chambers); enlarged aortic valve, sitting on top of VSD and opening from both ventricles as opposed to one. Surgery soon after birth	<1-5	Cy C*
Transposition of the Greater Arteries (TGA) [1, 5, 6, 7, 8, 14]	Main pulmonary artery and aorta are switched (transposed) in position. Initial surgical intervention in the first month of life.	5	Cy C*
Truncus Arteriosus (TA) [5,6,14,17]	A single blood vessel exits the heart instead of the usual two (aorta and main pulmonary artery). Requires multiple surgeries, beginning in the first few months of life.	<1	Cy C*
Total Anomalous Pulmonary Venous Connection or Return (TAPVC or TAPVR) [5,6,7, 16]	Pulmonary veins do not connect to the left atrium but through an abnormal connection. Can be complete or partial. Requires surgery in infancy to repair the defect- timing depends on severity.	<1-1	Cy C*
Ventricular Septal Defect (VSD) [1,6,7,8,11, 15]	One or more holes between the two ventricles (lower chambers of the heart). Treatment depends on the size and location of the hole/s. Small holes often self-correct over time. Other VSDs require cardiac catherization or surgery. Timing depends on severity.	20-42	A S

Classification: Acyanotic (A); Cyanotic (Cy); Simple (S); Complex(C); Critical (*)

[1]Van der Linde et al., 2011 [2]Fruitman, 2000 [3]Calabro & Limongelli, 2006 [4]Medline, 2016 [5]CDC, 2017 [6]American Heart Association, 2017 [7]NHS, 2015 [8]Saenz, Beebe & Triplett, 1999 [9]NIH, 2011 [10]Marelli, Mackie, Ionescu-Ittu, Rahme & Pilote, 2007 [11]Children's Heart Centre, 2017 [12] Western Australia Health, 2015 [13]American Heart Association, 2017 [14]Parker et al., 2010 [15]Reller, Strickland, Riehle-Colarusso, Mahle & Correa, 2008 [16]Bjornard, Riehle-Colarusso, Gilboa & Correa, 2013 [17]Royal Children's Hospital Melbourne, 2017 [18]Obler, Juraszek, Smoot & Natowicz, 2008 [19]Attenhofer Jost, Connolly, Dearani, Edwards & Danielson, 2007

For patients living with some of the most complex and life-threatening forms of CHD, such as HLHS, recent medical and surgical advancements have occurred at such rates that life-saving surgeries not available at the time of an infant's birth, may become available by the time the developing child requires them (Gordon, Rodriguez, Lee & Chang, 2008; Maher, Gidding, Baffa, Pizarro & Norwood, 2004). For patients and their families this offers hope, but also a pervasive experience of uncertainty and constant threat of the child's death (Rempel, Rogers, Ravindran & Magill-Evans, 2012). Whilst these increases in surgical interventions are responsible for increased longevity and survival, learnings from oncology research suggest that surgical and other medical interventions also impact psychologically on both the child and their families and can have adverse implications for a family's ability to cope (e.g. Angstrom-Brannstrom, Norberg, Strandberg, Soderberg & Dahlqvist, 2010; Bayat, Erdem & Gul Kuzucu, 2008; Kazak, 2005; Kazak et al., 1997; Twombly, 2001).

1.1.3 Coping and psychological distress in families of children with CHD

Three systematic literature reviews explored the experience of psychological distress and coping in families who have a child with CHD (Jackson, Frydenberg, Liang, Higgins & Murphy, 2015; Soulvie, Desai, White & Sullivan, 2012; Wei, Roscigno, Hanson & Swanson, 2015). Whilst each of these studies identified common research themes in the literature, inconsistencies and gaps in research findings were also found. In 2012, Soulvie et al. reviewed literature regarding psychological distress in parents of young children (aged 0-5) with CHD (n=25). The review found increased prevalence of stress (n=9); worry and concern (n=5); anxiety and depression (n=9); and other distressing emotions (fear, anger, helplessness, guilt, powerlessness and disbelief; n=3) in parents of children with congenital heart disease (PCCHD), as compared to normative populations. Further, three major categories of stressors were identified: diagnosis and treatment; parenting during the child's illness; and impact on intrafamilial relationships. Research gaps included: longitudinal

studies of distress (prenatal onwards); mixed design studies indicating good integration of qualitative and quantitative findings; groups of studies using consistent measures of distress; exploration of guilt and grief; studies of parental depression and anxiety beyond infancy; and exploration of the concept of uncertainty.

In a 2015 review of families of children with CHD (n=94), Wei et al. identified four themes: parents' psychological health (n=24); family life (n=21); parenting challenges (n=18); and family-focused interventions (n=5). Inconsistencies were found in relation to parental psychological health, with 75% of studies showing levels lower than normative samples and 25% showing levels commensurate with, or higher than, the general population. Inconsistencies were attributed, in part, to the varied measures of psychological symptoms utilised. Gaps in the literature included a disproportionate number of studies on parenting challenges using samples of HLHS families (higher care needs); lack of studies using consistent measures of psychological distress; and reduced number of qualitative studies (25%) that may provide context not found in pre-programmed quantitative responses.

A systematic review undertaken in 2015 by Jackson et al. on the familial impact and coping of families of children with CHD (n=25) identified three major themes: impact on family; coping; and parenting. These studies consistently reported that CHD complexity was associated with increased parental distress and family uncertainty; and that PCCHD had increased severity and incidence of anxiety, distress, depression, hopelessness, somatisation and/or anger than normative samples. Whilst a variety of symptom clusters and diagnoses were identified under the broad construct of psychological distress within these reviews, traumatic stress was not identified as a diagnosis nor theme.

1.2 Trauma

Research into the effects of trauma in adult populations includes studies of post-traumatic stress (PTS) among returned armed-service personnel (e.g. Wisco et al., 2017);

emergency service personnel (e.g. Shakespeare-Finch, Smith & Obst, 2002; Wilson, 2015); those exposed to terrorist attacks (e.g. Abbas, Hassan & Ali, 2017; Wilson, 2015); and victims of crime (e.g. Ghafoori, Hansen, Garibay & Korosteleva, 2017; Shercliffe & Colotta, 2009), disasters (e.g. Arcaya et al., 2017; Heid, Christman, Pruchno, Cartwright & Wilson-Genderson, 2016; La Greca, Danzi & Chan, 2017) and accidents (e.g. Shercliffe & Colotta, 2009; Undavalli, Das, Dutt, Bhoi & Kashyap, 2014). In general, pre-trauma risk factors (such as gender, age, race, education, previous trauma and general childhood adversity) appear to be better predictors of Post-Traumatic Stress Disorder (PTSD) within specific trauma populations than in the overall population (Brewin, Andrews & Valentine, 2000). However, factors such as trauma severity, lack of social support and 'life stress' (factors that tend to operate during and post-trauma) are more generalisable predictors of PTSD in adult populations (Brewin et al., 2000).

1.2.1 Parental trauma

Parents have been shown to develop clinical levels of Post-Traumatic Stress Symptoms (PTSS), PTSD, and Acute Stress Disorder (ASD) when exposed to child-related trauma, such as a child's death (e.g. Applebaum & Burns, 2010; Murphy et al., 1999); disability (LeGouez et al., 2016; Roberts, Koenen, Lyall, Ascherio & Weisskopf, 2014); non-fatal accident (de Vries et al., 1999); abuse (Cyr et al., 2016); or chronic illness and/or medical interventions (e.g. Balluffi et al., 2004; Kazak et al., 2004). Prevalence rates vary considerably, with reports of 21-69% of mothers and 14-46% of fathers experiencing PTSD in relation to child-related trauma (Applebaum & Burns, 2010; de Vries et al., 1999; LeGouez et al., 2016; Murphy et al., 1999), and 32%-54% of parents meeting criteria for ASD (Baluffi et al., 2004; Muscara et al., 2015). In a longitudinal study of PTSD among bereaved parents of children (aged 12-28) who had died violent deaths, Murphy et al. (1999) reported that 21% of mothers and 14% of fathers showed persistent PTSD over a two-year

period. In addition, of the 32% of parents who met diagnostic criteria for ASD during their child's Pediatric Intensive Care Unit (PICU) admission, 21% progressed to PTSD at follow-up (> two months post-discharge; Baluffi et al., 2004). Further, Greene et al. (2015) identified PTS in parents of low birth-weight infants admitted to NICU (Neonatal Intensive Care Unit) and found that symptoms persisted for the duration of the admission, despite a decline in other symptoms of distress. The comorbidity of PTS with other symptoms of psychological distress was also found in a study of parents of children admitted to PICU with various life-threatening illnesses/injuries, where 49-54% met criteria for ASD, 15-27% for depression and anxiety, and 25-31% for general stress (Muscara et al., 2015). Ratings of distress (ASD, stress, anxiety and depression) did not differ significantly based upon diagnosis type, however ASD was significantly correlated with anxiety (r=.56), stress (r=.52) and depression (r=.49; Muscara et al., 2015).

1.2.2 Parental trauma and pediatric oncological diagnosis and treatment

Much of the research investigating parental experiences of child-disease-related-trauma is found within oncology. A systematic literature review in 2012, identified that parents of pediatric cancer patients are at risk of psychosocial morbidity, including symptoms of PTS such as intrusive thoughts, physiological responses, flashbacks and psychological agitation (Kohlsdorf & Costa Jnr). In general, research suggests that parents are at greater risk of PTSS as compared to normative samples (Houskamp & Kazak, 1996; Kazak et al., 2004 & 2005, Pelcovitz et al., 1996; Stuber, Christakis, Karadeniz et al., 2017; Ljungman, Hoven, Ljungman, Cernvall, & von Essen, 2015). However, there is inconsistency within the literature regarding prevalence (Kohlsdorf & Costa Jnr, 2012). Prevalence rates for PTSD have been shown to be between 21.7-30% (Karadeniz et al., 2017; Kazak et al., 2004 & 2005), with PTSS ranging from 33.3-99% (Kazak et al., 2004 & 2005; Stuber et al., 1996). Long term PTSD has also been identified in 13.7% of parents immediately following

completion of their child's treatment (Kazak et al., 2004) and in 8% fathers and 14% mothers five years later (Ljungman et al., 2015). Rates for bereaved parents at five-year follow up ranged from 20% for fathers and 35% for mothers (Ljungman et al., 2015). Lifetime PTSD rates are higher for parents of paediatric cancer survivors than for parents of healthy children (PHC. Pelcovitz et al., 1996).

Of interest, the trajectory of PTSS, PTSD and "generic distress" (depression and anxiety) in parents of pediatric cancer patients, has also been found to vary as a function of the time from initial diagnosis (Ljungman et al., 2015; Norberg & Boman, 2009). More specifically, Ljungman et al. (2015) found that whilst PTSS declined in the first months post-diagnosis, residual symptoms of PTSS (8% fathers and 19% mothers) became chronic in nature and were stable at five years post-treatment. Similarly, Norberg and Boman (2009) indicated that whilst the first 2.5 years post-diagnosis were characterised by severe generic distress, initial PTSS (such as arousal, avoidance and intrusive thoughts) became more severe and chronic in nature, with 12% of parents meeting diagnostic criteria for PTSD at the five-year follow-up and at levels commensurate with, or higher than, a clinical PTSD population.

1.3 Post-Traumatic Growth

Research into the experience of trauma in families of children with significant health concerns has tended to concentrate on psychopathology and focus on life-limiting conditions such as cancer (e.g. Angstrom-Brannstrom et al., 2010; Bayat et al., 2008; Clarke, McCarthy, Downie, Ashley & Anderson, 2009). More recently, however, research has shifted to include the experience of post-traumatic growth (PTG. e.g. Hensler, Catz, Wiener, Berkow & Madan-Swain, 2014; Hungerbuehler, Vollrath & Landolt, 2011). PTG can be defined as a positive psychological response to the experience of trauma, in which an individual not only recovers a previous level of functioning but is able to transcend this in some way, for example through increased spiritual connection or existential comprehension; sense of stronger self; re-

prioritisation of what is considered important in life; and/or more meaningful relationships with others (Calhoun & Tedeschi, 1999, 2001; Tedeschi & Calhoun, 1996, 2004). Resilience, which has been defined as the ability to maintain a previous level of functioning during and after adversity (Werner, 1995) and, more recently, as positive adaptation in the face of adversity (Luthar, 2006), shares some features with PTG, however PTG is generally understood to be a distinct construct in that it extends recovery beyond coping or adaptation into psychological growth, 'meaning finding' or 'thriving' (Carver, 1998; Tedeschi & Calhoun, 2004).

Further, research suggests that whilst PTG and PTS are distinct constructs with separate predictors, they are related and co-occurrence is common (Armstrong, Shakespeare-Finch & Shochet, 2014; Shakespeare-Finch & Lurie-Beck, 2014; Tedeschi & Calhoun, 2004). The extent to which PTG and PTS co-occur varies based on trauma type and age (Shakespeare-Finch & Lurie-Beck, 2014). In a 2014 meta-analysis (n=11,469), PTG and symptoms of PTSD were shown to co-occur to such a degree that Shakespeare-Finch and Lurie-Beck cautioned that focusing solely on psychopathology may not only 'mask' growth but also have adverse consequences for recovery.

PTG research has been applied to a variety of fields (Shakespeare-Finch & Beck) and has also been identified in parents of children who have experienced life-limiting or life-threatening medical concerns (e.g. Turner-Sack, Menna, Setchell, Maan & Cataudella, 2016). PTG has been found in 54.3% parents of children undergoing corrective surgery for congenital defects (Li, Cao, Cao, Wang & Cui, 2012). In the same study, Li et al. (2012) found that PTG was positively related to resilience, self-efficacy and emotional intelligence, along with PTSS. In addition, it was found that whilst emotional intelligence was the main predictor for PTG, resilience and PTSS mediated emotional intelligence, along with self-efficacy, perceived social support and growth.

1.4 Scope of the Current Review

This systematic literature review is designed to explore the experience of psychological distress, trauma-related symptomology, coping and PTG among parents of children with CHD. Acknowledging Ridner's (2004) finding that the term psychological distress constitutes a variety of definitions, for purposes of this literature review, psychological distress is defined in the broadest of terms as 'emotional suffering' that impacts on the day-to-day functioning of an individual (Wheaton, 2007). 'Emotional suffering' is explored not only through the symptoms (and general measures) of mental ill-health and/or psychological distress (such as the Brief Symptom Inventory (BSI), General Health Questionnaire (GHQ), Mental Health Inventory (MHI), Parenting Stress Index (PSI) and Symptom Checklist (SCL)), and also through specific diagnostic or symptom clusters such as depression, anxiety, stress and somatisation (Kirmayer, 1989; Kleinman, 1991; Mirowsky & Ross, 2002).

Trauma-related symptomology (or PTSS) is defined as those physical, cognitive, behavioural and emotional symptoms commonly experienced as part of conditions such as PTSD, Adjustment Disorder and ASD (APS, 2017). Coping is defined broadly to include the mechanisms that parents employ in order to attempt to psychologically adjust to their child's CHD and to maintain functioning (Lazarus & Folkman, 1984). This definition includes both maladaptive and adaptive (or resiliency building) strategies (Luthar, 2006). Consistent with Tedeschi & Calhoun (2004), PTG is defined as the manner in which parents find ways to build upon pre-trauma functioning through such practices as increased spiritual connection or existential comprehension; sense of stronger self; re-prioritisation of what is considered important in life; and more meaningful relationships with others.

1.5 Expected Benefits of the Current Study

The current study has a number of expected benefits. First, it adds to current literature about the psychological impact for PCCHD and builds upon previous reviews and meta-analyses within this field (the most recent of which was two years ago. Jackson et al., 2015; Soulvie et al., 2012; Wei et al., 2015). By applying broad research definitions to include a range of studies, the current review responds to the recommendations of Soulvie et al. (2012), specifically to reduce the limitation of narrow search constructs; explore psychological distress beyond infancy to older ages; identify the existence of wider symptoms of distress; and integrate the findings of quantitative and qualitative studies. In addition, the inclusion of a specific exploration of PTSS and PTG is, to the author's best knowledge, a novel contribution to reviews within this field.

Second, research suggests that reduced child psychological and medical outcomes are linked to poorer parental mental health (Lambert, Holzer & Hasburn, 2014; Melnyk et al., 2004). More specifically, a 2014 meta-analysis identified that increased severity of parental PTSD was positively correlated with levels of psychological distress in children (r=.35), regardless of trauma context or whether it was experienced by the parent alone or by the parent-child dyad (Lambert et al., 2014). Specifically in relation to CHD, Goldberg, Morris, Simmons, Fowler & Levison (1990) found a positive relationship between maternal attachment style and mental health, and improvements in physical health for securely attached infants. The potential benefits for improving the physical and psychological health outcomes for children with CHD by better understanding, and responding to, the psychological reactions of their parents are clear (e.g. children that are less distressed, more resilient and physically stronger).

Third, within Australia, mental ill-health has been identified as the largest contributor to non-fatal disability burden, with an estimated cost of $20 billion per annum (inclusive of

productivity loss and decreased participation in work ABS, 2013; AIHW, 2007). In addition, acute coronary episodes are estimated to cost $18.3 billion per annum (financial burden of $5billion and burden of disease cost of $13.3 billion), and chronic stress is a contributor to multiple physical conditions, including increased risk for hypertension, heart attack and stroke (APA, 2013; Deloitte, 2011). This study will also contribute to an understanding of the prevalence, development and trajectory of mental ill-health in parents of children with CHD, and the ways in which this may be best prevented or mitigated, to reduce burden on mental-health and medical services for these parents into the future. Taken together, this review is anticipated to contribute to the research base available to guide the development of public health policy and intervention programs, and to identify research gaps necessitating future exploration.

Chapter 2 - Method

2.1 Aim and Research Questions

The aim of this review was to investigate the experience of psychological distress, PTS, PTG and coping in PCCHD. This aim was operationalised as 10 research questions (Table 2.1).

Table 2.1

Research Questions

Number	Question
RQ1	Do PCCHD experience psychological distress and what forms does this take?
RQ2	Do PCCHD experience psychological distress that is greater or similar to that of parents of healthy children and parents of children with other disorders?
RQ3	Does prenatal versus postnatal diagnosis impact on the psychological distress experienced by PCCHD?
RQ4	Is psychological distress of PCCHD related to the severity or diagnostic category of cardiac condition, or to the requirement for (or stage of) surgery?
RQ5	Does psychological distress in PCCHD change over time?
RQ6	Are there differences in psychological distress based upon the gender of the PCCHD?
RQ7	Do PCCHD experience clinical levels of trauma (Acute Stress Disorder, PTSD, PTSS)?
RQ8	Do PCCHD experience PTG?
RQ9	What coping strategies are employed by PCCHD and which are most successful in reducing psychological distress or clinical levels of trauma?
RQ10	What interventions exist to reduce psychological distress in PCCHD and are they effective?

2.2 Procedure

Research protocol and parameters were defined and submitted for registration review on PROSPERO. PRISMA guidelines were followed for the identification and extraction of articles for inclusion (Liberati et al., 2009). EBSCOhost, Illumina, JSTOR and PubMed databases were searched by entering the Boolean search string "(PTSD OR post-traumatic stress disorder OR post traumatic stress disorder OR posttraumatic stress disorder OR PTSS OR post-traumatic stress symptoms OR post traumatic stress symptoms OR posttraumatic stress symptoms OR PTG OR post-traumatic growth OR post traumatic growth OR post

traumatic growth OR distress OR anxiety OR depression OR psychosocial OR quality of life OR coping OR acute stress disorder OR adjustment disorder) AND (CHD OR cardiac OR heart) AND (Paren* OR mothe* OR fathe* OR patern* OR matern*)". Searches were limited to the titles and abstracts of peer-reviewed scholarly articles in English which had full-text readily available online. Search strings were developed through consultation with the author's supervisor, research students and the university librarian, and in consideration of specific research questions.

An initial 2,607 articles were found and imported into endnote X7, with 1654 of these removed as duplicates. A screening of titles and abstracts was undertaken based on pre-specified inclusion and exclusion criteria (Table 2.2) resulting in the removal of 782 articles. A detailed examination of remaining articles was then undertaken based on the inclusion and exclusion criteria. An additional 46 articles were identified for consideration through suggested citations and examination of reference lists from selected articles and existing reviews (Soulvie et al., 2012; Wei et al., 2015; Jackson et al., 2015). Where full-text articles were not available online, an attempt was made to contact authors to request these. Thus, ninety-four articles were retained for quality review. (Figure 1.1).

Table 2.2

Inclusion, Exclusion and Special Consideration Criteria

Type of Criteria	Code	Details
Inclusion	I1	Foetus, infant or child with CHD (prenatal up to 18 years)
	I2	Research question or measure or qualitative theme of parent psychological distress, traumatic-stress, coping (including resilience) or PTG
	I3	Full text available
	I4	English
	I5	Peer-refereed journal
	I6	Relevant to research question/s

Exclusion criteria	E1	N=1 studies, single case study
	E2	Literature review
	E3	Program description where no relevant parent outcome measures were undertaken
	E4	Prenatal studies focused on counselling for termination decisions
	E6	End of life, palliative, bereavement studies
	E7	Quality rating below 5 (Low to Very Low grade of recommendation)
Special consideration criteria	SC1	Studies of mixed child illness - data on CHD parents extractable or 80%
	SC2	Studies of mixed caregivers - data on CHD parents extractable or 80%
	SC3	Studies where child disease/disorder includes CHD as a component but where other significant impairments exist (e.g. Downs Syndrome) - focus of study is on CHD
	SC4	Studies of mixed foetal/child age where range extends beyond 18 - data on CHD minors is extractable or 80% or mean age is under 18 (and range extends no more than 25)

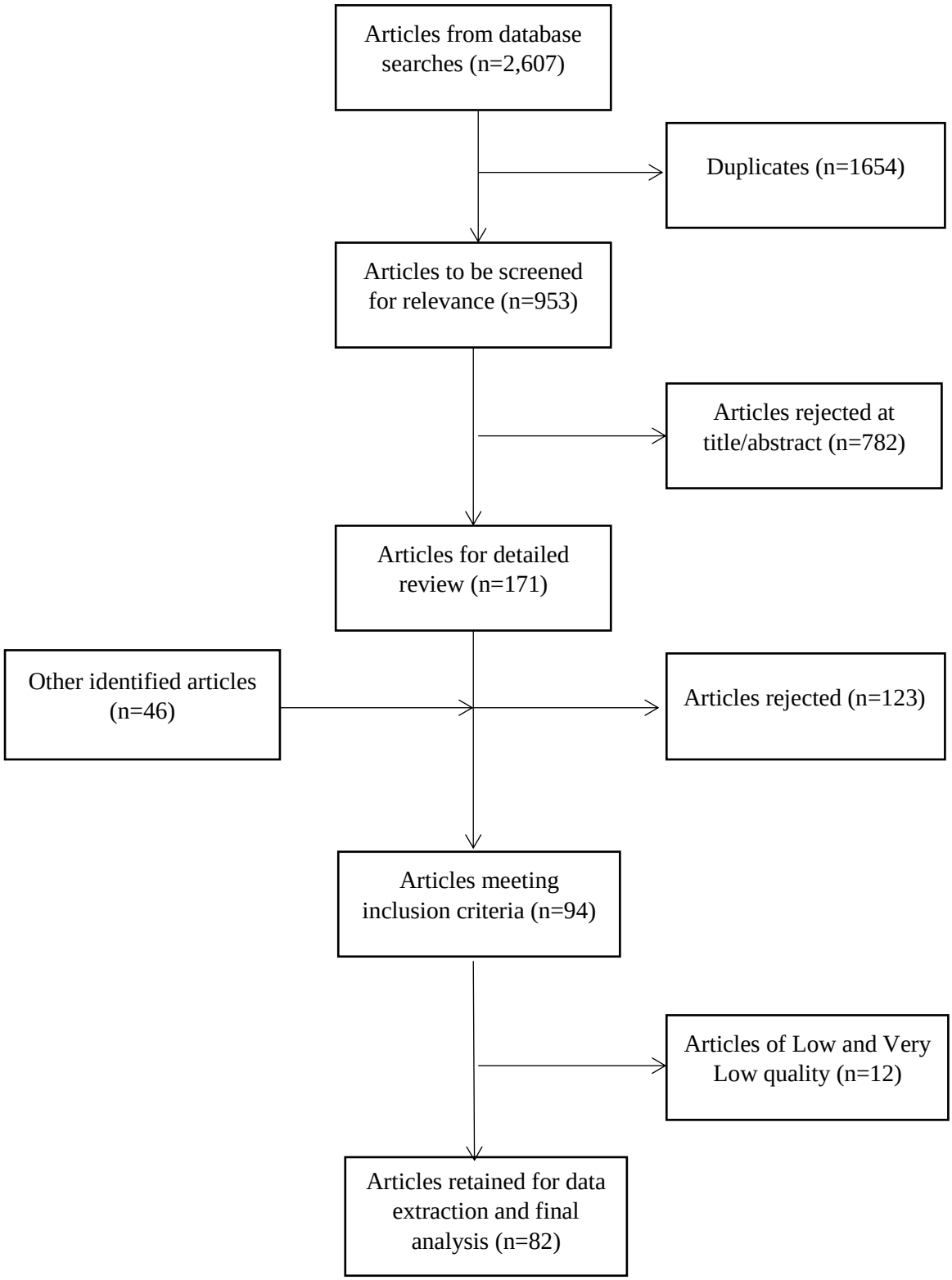

Figure 1: PRISMA flow-chart for article selection

2.3 Article Selection and Quality Ratings

The author evaluated articles against the inclusion and exclusion criteria to select those articles for quality review. Article selection and the assessment of quality was undertaken by the author and then verified by a colleague with qualifications in psychology and experience within the field of mental health and trauma. Where discrepancy existed, the case was discussed with the author's university supervisor (Clinical and Health Psychologist and researcher) until consensus was reached. Of the 94 articles considered for this review, 12 were excluded based on their quality rating (exclusion criteria E7). Of the 82 included within this review, 11 were retained based upon special consideration criteria (Table 2.3).

Table 2.3

Articles Retained Based Upon Special Consideration Criteria

Reference	Criteria	Reason for consideration
Arafa et al. (2008)	SC1	Examination of heart disease (67.5% CHD, 32.5% Rheumatic heart disease). Some data separated by type of heart disease (CHD versus RHD).
Goldbeck & Melches (2006)	SC2; SC4	'Children' included young adults, however mean age was 7.8. Parents were 99% of the sample, 1% were other primary caregivers.
Gudmundsdottir et al., (1996)	SC4	Age range of 'children' was 13-25 years old. 62.5 <18 years, however mean was 17.75 years.
Lawoko & Soares, (2002)	SC4	'Children' were aged 0-20 years, however mean was 7.2.
Lawoko & Soares, (2003a)	SC4	'Children' were aged 0-20 years, however mean was 7.2.
Lawoko & Soares, (2003b)	SC4	'Children' were aged 0-20 years, however mean was 7.2.
Lawoko & Soares, (2006)	SC4	'Children' were aged 0-20 years, however mean was 8 and all were cared for in the family home.
Levert et al. (2017)	SC3	Children had other comorbidities, however the focus was on CHD
McCusker et al. (2010)	SC3	Study included 20% children with Downs Syndrome and 10% with other developmental delay, however all had CHD and the focus of the study was on CHD.
Sira et al. (2014)	SC4	'Children' included those that were >18 years, however this was only 8.6% of the sample. No upper age limit was provided. A decision was made to include the study based upon the specific and unique focus upon spirituality and

		internet based social connections, and consideration of these under definitions of PTG
Sparacino et al. (1997)	SC4	'Child' age ranged from 13-25 years and 37.5% were over 18, however mean age was <18 years

A modified version of the methodology used by Ireland et al. (2017) was used for critically appraising and rating both the level of evidence and quality of studies included in this review. Specifically, each study was assigned a level of evidence; quality rating; and quality score based on existing frameworks for critical appraisal of quality (Table 2.4). In keeping with Ireland et al., existing appraisal frameworks were selected based on their applicability to the design of studies under review and their evidence base. As this review included qualitative, quantitative and mixed design studies (with varying methodologies), a variety of approaches were selected.

This review included a large proportion of cohort and cross-sectional studies. In keeping with Ireland et al. (2017), non-randomised quantitative studies were assessed using the Newcastle-Ottawa Scale (NOS) checklists (for cohort studies; Wells et al., 2000. Appendix B) and the NOS modified version (for cross-sectional studies; e.g. Modesti et al., 2016. Appendix C). The NOS is a reliable and valid measure specifically designed and successfully used in reviews of non-randomised designs (Chen et al., 2010; Wells et al., 2011; Young et al., 2011). The approach uses a weighted checklist, with the total score (1 the lowest and 9 or 10 the highest depending on the checklist used) derived from three subscales (selection, comparability and outcomes). The Oxford Centre for Evidence Based Medicine (OCEBM) methodology was used to derive levels of evidence for each study (Appendix D). The OCEBM Levels of Evidence (version 2) uses a matrix approach to assign values to the methodology of each study (I the highest, IV the lowest) based on the purpose of the study and is widely utilised in health-related research reviews (Cooper, Balamurali & Livingstone, 2007; Howick et al., 2011). It is

similar to the NHMRC model (used by Ireland et al., 2017; NHMRC, 2009) but has a stronger focus on prevalence studies (more relevant to the studies selected for this review).

Qualitative studies were assessed using the JBI model (Pearson, Wiechular, Court & Lockwood, 2005) which evaluates the quality of studies based on their purpose (Levels of Evidence) and methodology (Critical Appraisal Checklists). The JBI method has been successfully used in a number of psychological-health-related systematic reviews (e.g. Townshend, Jordan, Stephenson & Tsey, 2016) and provides critical appraisal checklists based on study methodology, as well as levels of evidence based on study context. Unlike many appraisal methods, it allows for the evaluation of qualitative studies and has been used successfully within this context (e.g. Fawcett, Porritt, Campbell & Carson, 2017; Suleman, Manning & Evans, 2016). The JBI Checklist for Qualitative Studies was completed and scored for each applicable study (score out of 10 with 10 the highest. Appendix E) and the JBI Level of Evidence for Meaning (with 1 the highest and 5 the lowest) was applied based on study methodology (Appendix F).

This review included studies of a mixed methods design. Consistent with the approach of Ireland et al. (2017), these were each assigned as being qualitative or quantitative based upon their predominant approach/research question, and were then critically appraised within this context.

This study also included a very small number of quantitative studies with randomised control or test accuracy designs (n=4). As the Newcastle-Ottawa Scales were not applicable to these quantitative designs, the JBI approach was applied in these cases: Checklist for RCT (score out of 13 with 13 the highest); Checklist for Diagnostic Accuracy (out of 10); Level of Evidence for Effectiveness (1a the highest, 5c the lowest); and Level of Evidence for Diagnosis (1a the highest, 5c the lowest. Appendices G-J).

For comparison across studies (consistent with Ireland et al., 2017), the critical appraisal score was converted to a score out of 10 (quality score) for each study. A four-point scale was then applied to the quality score to determine the quality ratings. Scores of 0 to 2.49 were Very Low; 2.5 to 4.99 were Low; 5-7.49 were Moderate; and 7.5-10 were High. Consistent with Townshend et al., (2016), studies below 5 (Very Low or Low quality) were excluded from this review (Figure 1). Of the 82 studies retained, over three quarters were rated as High quality (n=65). Quality scores, quality ratings and levels of evidence can be found in Table 2.3.

2.4 Data Extraction

Data extraction was undertaken by the author and verified by a colleague within the field. Articles were first matched against the 10 research questions (Table 2.1) and data was then extracted against selected fields within an Excel version 10 spreadsheet (sample size, parent gender, foetus/child age, CHD diagnosis, CHD severity, surgical requirement/status, study design, measures used, country of origin of study, major related findings).

During detailed examination of selected articles, a memo style approach (consistent with the methodology of Wei et al., 2015) was utilised to identify themes related to the research questions as they emerged (e.g. specific symptom clusters such as depression, anxiety, somatisation and stress). These themes were then entered as fields and the process of data extraction was repeated. Table K1 presents data and major findings for each study (Appendix K).

2.5 Comparison with Existing Literature Reviews

Studies within the three existing reviews (Soulvie et al., 2012; Wei et al., 2015; Jackson et al., 2015) were cross-referenced against articles selected for the current review to ensure the unique contribution of this review (combined 120 studies). Of the 82 articles

selected for this review, 46 were within at least one other review, 4 were within all reviews and 36 were unique to this review (Table L1, Appendix L).

2.6 Structure of Results

Research questions within this review are inter-related. More specifically, RQs 2-6 (e.g. the impact of CHD severity, diagnoses and surgery) are applicable to each diagnostic category identified through investigation of RQ1 (what forms does psychological distress take?). To improve cohesion and readability, results have been structured by diagnostic category and then broken down by sub-themes (Table 2.5). Trends in sub-themes common between diagnoses are explored in the discussion.

Table 2.5

Research Questions Mapped by Review Sub-Sections

Subsection	Research Question
Psychological Distress, Stress, Depression, Anxiety, Somatisation	1,2,3,4,5,6,10
Prevalence	1
Comparison to PHC and PCOD	2
Impact of timing of CHD diagnosis	3
Impact of CHD severity	4
Impact of surgery	5
Influence of parent gender	6
Interventions	10
Trauma	7
Clinically Significant Trauma	7
Evidence of Trauma Symptomology	7
Post-Traumatic Growth	8
Coping and Resilience	9, 10
Comparison of coping in PCCHD to PHC and PCOD	9
CHD severity and coping	9
Influence of parent gender and coping	9
Coping strategies used	9
Helpful coping strategies	9
Unhelpful coping strategies	9
Interventions and coping	10

Table 2.4

Study Types. Quality Ratings, Grades of Recommendation and Levels of Evidence

Author (date)	Type	Checklist	Quality Rating (/10)	Grade of recommendation	JBI Level of evidence
Bratt et al. (2015)^^^	Qual	JBI	9	High	3
Bruce & Sundin (2012)^^^	Qual	JBI	9	High	3
Bruce et al. (2014)^^^	Qual	JBI	8	High	3
Bruce et al. (2016)^^^	Qual	JBI	9	High	3
Cantwell-Bartl & Tibballs (2015)^^^	Qual	JBI	8	High	3
Clark & Miles (1999)^^^	Qual	JBI	8	High	3
Connor et al. (2010)^^^	Qual	JBI	9	High	3
Gudmundsdottir et al. (1996)^^^	Qual	JBI	10	High	3
Harvey et al. (2013)^^^	Qual	JBI	9	High	3
Kocyildirim et al. (2007)^^^	Qual	JBI	7	Moderate	3
Kosta et al. (2015)^^^	Qual	JBI	9	High	3
Lan et al. (2007)^^^	Qual	JBI	10	High	3
Lee & Rempel (2011)^^^	Qual	JBI	7	Moderate	3

Lok & Menahem (2004)^^^	Qual	JBI	8	High	3
Meakins et al. (2015)^^^	Qual	JBI	9	High	2
Pinelli (1981)^^^	Qual	JBI	7	Moderate	3
Pridham et al. (2010)^^^	Qual	JBI	7	Moderate	3
Redshaw et al. (2011)^^^	Qual	JBI	10	High	3
Rempel & Harrison. (2007)^^^	Qual	JBI	10	High	3
Rempel et al. (2009)^^^	Qual	JBI	10	High	3
Sparacino et al. (1997)^^^	Qual	JBI	9	High	3
White et al. (2016)^^^	Qual	JBI	9	High	3
					CEBM level of evidence
Arafa et al. (2008)*	Quan	NOS	7.8	High	2
Berant et al. (2001)**	Quan	NOS	6.7	Moderate	2
Berant et al. (2003)**	Quan	NOS	6.7	Moderate	2
Berant et al. (2008)**	Quan	NOS	6.7	Moderate	2
Bevilacqua et al. (2013)**	Quan	NOS	10	High	2
Brosig, Mussato et al. (2007)*	Quan	NOS	6.7	Moderate	2

Davis et al. (1998)*	Quan	NOS	7.8	High	2
Diffin et al. (2016)**	Quan	NOS	8.9	High	2
Doherty et al. (2009)*	Quan	NOS	6.7	Moderate	2
Fischer et al. (2012)*	Quan	NOS	6.7	Moderate	2
Franich-Ray et al. (2013)*	Quan	NOS	10	High	2
Goldbeck & Melches (2005)*	Quan	NOS	10	High	2
Goldbeck & Melches (2006)*	Quan	NOS	8.9	High	2
Gronning-Dale et al. (2012)**	Quan	NOS	10	High	2
Gronning-Dale et al. (2013)**	Quan	NOS	10	High	2
Hearps et al. (2014)*	Quan	NOS	10	High	2
Landolt (2011)**	Quan	NOS	8.9	High	2
Lawoko & Soares (2002)*	Quan	NOS	10	High	2
Lawoko & Soares (2003a)*	Quan	NOS	10	High	2
Lawoko & Soares (2003b)*	Quan	NOS	10	High	2
Lawoko & Soares (2004)*	Quan	NOS	10	High	2
Lawoko & Soares (2006)**	Quan	NOS	8.9	High	3

Lee et al. (2007)*	Quan	NOS	7.8	High	2
Levert et al. (2017)*	Quan	NOS	6.7	Moderate	2
Majnemer et al. (2006)**	Quan	NOS	8.9	High	2
Pelchat et al. (1999)*	Quan	NOS	8.9	High	2
Pinto et al. (2016)**	Quan	NOS	10	High	2
Rahimianafar et al. (2015)*	Quan	NOS	6.7	Moderate	3
Rona et al. (1998)**	Quan	NOS	10	High	2
Rychik et al. (2013)*	Quan	NOS	10	High	2
Sarajuuri et al. (2012)**	Quan	NOS	10	High	2
Sira et al. (2014)*	Quan	NOS	10	High	2
Sklansky et al. (2002)**	Quan	NOS	7.8	High	3
Solberg et al. (2011a)**	Quan	NOS	8.9	High	3
Solberg et al. (2011b)**	Quan	NOS	10	High	3
Solberg et al. (2012)**	Quan	NOS	10	High	3
Spijkerboer et al. (2007)**	Quan	NOS	10	High	2
Svavarsdottir & McCubbin (1996)*	Quan	NOS	7.8	High	4

Tak & McCubbin (2002)*	Quan	NOS	8.9	High	3
Torowitz et al. (2010)*	Quan	NOS	7.8	High	3
Utens et al. (2002)**	Quan	NOS	8.9	High	2
Uzark & Jones (2003)*	Quan	NOS	7.8	High	3
Vrijmoet-Wiersma et al. (2009)*	Quan	NOS	7.8	High	2
Werner et al. (2014)**	Quan	NOS	6.7	Moderate	3
Wilson & Chando (2015)*	Quan	NOS	6.7	Moderate	4
Wray & Sensky (2004)**	Quan	NOS	8.9	High	2
Yildiz et al. (2009)*	Quan	NOS	7.8	High	3
					JBI level of evidence
Dulfer et al. (2015)^	Quan	JBI	6.9	Moderate	1c
Helfricht et al. (2009)^^	Quan	JBI	10	High	1b
McCusker et al. (2010)^	Quan	JBI	7.7	High	1d
McCusker et al. (2012)^	Quan	JBI	8.5	High	1c
					CEBM level of evidence
Brosig, Whitstone et al. (2007)**	Mixed design	NOS	8.9	High	2

Franck et al. (2010)**	Mixed design	NOS	8.9	High	3
Gardner et al. (1996)**	Mixed design	NOS	8.9	High	2
Hoehn et al. (2004)*	Mixed design	NOS	8.9	High	2
Jordan et al. (2014)*	Mixed design	NOS	10	High	2
Odegard et al. (2002)*	Mixed design	NOS	5.6	Moderate	4
					JBI level of evidence
Bright et al (2013)[1]	Mixed design	JBI	8	High	2
Carey et al (2002)[1]	Mixed design	JBI	8	High	2
McCrossan et al (2008)[2]	Mixed design	JBI	5.6	Moderate	2d

*NOS - Cross-sectional
**NOS - Cohort
^JBI RCT Checklist
^^JBI Diagnostic Test Accuracy Checklist
^^^JBI Qualitative Checklist
[1]JBI Qualitative Checklist
[2]JBI Quasi-Experimental Checklist

Chapter 3 - Results

3.1 Descriptive Statistics

Eighty-two studies were selected for inclusion. Of these 51 were quantitative (62.19%), 22 were qualitative (26.83%), and 9 were of mixed method design (10.98%). Sample sizes ranged from five to 61,511. Repeated measures were used in just over one third (n=31, 37.80%). The studies originated in 18 countries, with the highest frequency of studies in the United States (n=21, 25.61%), Australia (n=9, 10.98%), Sweden (n=9, 10.98%) and Canada (n=8, 9.76%).

Almost two thirds of studies involved parents of mixed gender (n= 54, 65.85%), with almost one third focused on mothers (n=25, 30.49%) and the remaining smaller number of studies on fathers (n=3, 3.66%). Most studies were concerned with either children of varied age (n=36, 43.90%) or neonates/infants (n=37, 42.68%). The remaining studies (13.41%) concerned adolescents (n=1), pre-schoolers (n=4), foetuses (n=4) and infant/foetuses (n=2).

The majority of studies included mixed CHD diagnoses (n=71, 86.59%), however 10 studies (12.20%) focused on SVP (with 7 specifying HLHS). Three quarters of the studies examined complex CHD (n=62, 75.61%). Over half of the studies exclusively focused on participants in families where the child required surgical intervention (including cardiac catheterisation, n=39, 47.56%), with 26 (31.71%) of these studies conducted exclusively post-surgery.

Seventy-three validated measures and two un-validated questionnaires were used in the quantitative/mixed design studies (n=60). The most frequent measures used were un-validated measures (n=7, 8.54%), STAI (n=7, 8.54%), and BSI (n=5, 6.10%). Multiple versions of the SCL (n=9, 10.98%), PSI (n=9, 10.98%) and GHQ (n=8, 9.76%) were also frequently used. Descriptive statistics are in Table 3.1. Frequency scores for measures can be found in Appendix M.

Table 3.1

Descriptive Statistics

Descriptive	N	Percentage of total studies
Study design		
Quan	51	62.19
Qual	22	26.83
Mixed	9	10.98
Repeated measures	31	
Parent Type		
Fathers	3	3.66
Mothers	25	30.49
Mixed	54	65.85
Child age		
Mixed	36	43.90
Foetus	4	4.88
Foetus/Infant	2	2.44
Neonate	15	18.29
Neonate/Infant	3	3.66
Infant	17	20.73
Pre-schooler	4	4.88
Adolescent	1	1.22
CHD Diagnoses		
Mixed	71	86.59
Specified	11	13.41
CHD Complexity		
Mixed	62	75.61
Complex	16	19.51
Moderate-to-severe	1	1.22
Simple-to-moderate	1	1.22
Surgical Status		
Mixed	36	43.90
Pre	11	13.41
Post	26	31.71
During	1	1.22
Country		
Australia	9	10.98
Canada	8	9.76
Egypt	1	1.22
Finland	1	1.22
Germany	3	3.66
Iran	1	1.22
Ireland	1	1.22
Israel	3	3.66
Italy	1	1.22
Korea	1	1.22
Netherlands	4	4.88
Northern Ireland	3	3.66
Norway	5	6.10
Sweden	9	10.98
Switzerland	3	3.66
Turkey	1	1.22
United Kingdom	6	7.32
United States	21	25.61

3.2 Non-Specific Psychological Distress (RQ1)

Non-specific psychological distress was discussed in 36 studies (mixed design n=4; quantitative n=24; qualitative n=8. Table 3.2).

Table 3.2

Studies Addressing Non-Specific Psychological Distress in PCCHD

Author (date)	Type	Topic	RQ
Arafa et al. (2008)	Quan	COD/HC comparison	2
Berant et al. (2001)	Quan	Severity, other factors	4
Berant et al. (2008)	Quan	Severity, other factors	4
Bevilacqua et al. (2013)	Quan	Prevalence, surgery, parent gender	1, 5, 6
Brosig, Whitstone et al. (2007)	Mixed design	Prevalence, COD/HC comparison, diagnosis, severity	1, 2, 3, 4
Cantwell-Bartl & Tibballs (2015)	Qual	General themes, diagnosis	1, 3
Carey et al (2002)	Mixed design	General themes	1
Clark & Miles (1999)	Qual	General themes	1
Connor et al. (2002)	Qual	General themes, diagnosis	1, 3
Davis et al. (1998)	Quan	Prevalence	1
Doherty et al. (2009)	Quan	Severity, surgery, parent gender, other factors	4, 5, 6
Dulfer et al. (2015)	Quan	COD/HC comparison	2
Gardner et al. (1996)	Mixed design	General themes, COD/HC comparison, surgery	1, 2, 5
Goldbeck & Melches (2005)	Quan	COD/HC comparison	2
Goldbeck & Melches (2006)	Quan	Severity	4
Gronning-Dale et al. (2012)	Quan	COD/HC comparison	2
Gronning-Dale et al. (2013)	Quan	COD/HC comparison, severity	2, 4

Hearps et al. (2014)	Quan	Prevalence, diagnosis, severity	1, 3, 4
Harvey et al. (2013)	Qual	General themes	1
Hoehn et al. (2004)	Mixed design	General themes	1
Kocyildirim et al. (2007)	Qual	General themes	1
Lan et al. (2007)	Qual	General themes	1
Lok & Menahem (2004)	Qual	General themes	1
Landolt (2011)	Quan	Surgery	5
Lawoko & Soares (2002)	Quan	COD/HC comparison, severity, parent gender, other factors	2, 4, 6
Lawoko & Soares (2003a)	Quan	COD/HC comparison, parent gender, other factors	2, 6
Lawoko & Soares (2004)	Quan	Severity	4
Levert et al. (2017)	Quan	Prevalence, other factors	1
McCusker et al. (2012)	Quan	Intervention	10
Meakins et al. (2015)	Qual	General themes	1
Pelchat et al. (1999)	Quan	COD/HC comparison	2
Pinto et al. (2016)	Quan	Diagnosis, gender	3, 6
Sklansky et al. (2002)	Quan	COD/HC comparison	2
Spijkerboer et al. (2007)	Quan	COD/HC comparison	2
Utens et al. (2002)	Quan	Surgery, other factors	5
Wray & Sensky (2004)	Quan	Prevalence, COD/HC comparison, severity, surgery, parent gender, other factors	1, 2, 4, 5, 6
Yildiz et al. (2009)	Quan	Severity, parent gender, other factors	4, 6

Qualitatively, four main themes of psychological distress in PCCHD were identified: distress, anguish and devastation (Cantwell-Bart & Tibballs, 2015; Connor, Kline, Mott, Harris & Jenkins, 2010; Gardner, Freeman, Black & Angelini, 1996; Harvey, Kovalesky, Woods &

Loan, 2013; Lok & Menahem, 2004); shock, puzzlement and disbelief (Carey, Nicholson & Fox, 2002; Harvey et al., 2013; Lan, Mu & Hsieh, 2007); loss of control and uncertainty (Cantwell-Bart & Tibballs, 2015; Clark & Miles, 1999; Connor et al., 2010; Harvey et al., 2013; Kocyilidrim, Franck & Elliott, 2007; Meakins et al., 2015); intense emotion, emotionally drained or overwhelmed (Clark & Miles, 1999; Connor et al., 2010; Harvey et al., 2013; Hoehn et al., 2004).

3.2.1 Prevalence of non-specific psychological distress (RQ1)

Levert, Helbing, Dulfer and van Domburg (2017), reported that 40% of PCCHD identified needing psychosocial support. Likewise, Hearps et al. (2014) found that 35.9% PCCHD required psychosocial intervention and a further 2.6% were at clinically significant psychosocial risk. Similarly, Davis, Brown, Bakeman and Campbell (1998) found that 37.3% of mothers met criteria for poor adjustment and Doherty et al. (2009) found that 18% of fathers, and 33% of mothers, had clinically significant mental health difficulties. Brosig, Whitstone, Frommelt, Frisbee and Leuthner (2007) found that at time of diagnosis 58% - 71% of women were distressed, with distress at time of birth ranging from 71% to 75%.

Bevilacqua et al. (2013) reported that 60-82% of parents whose infants had undergone cardiac surgery before four months, experienced psychological distress. In studies measuring pre-surgery distress for parents whose children required surgery for CHD, Wray and Sensky (2004) found psychological distress in 48-65% of PCCHD and Gardner et al. (1996) found clinically significant levels in 75% of mothers.

3.2.2 Comparison of psychological distress in PCCHD to PCOD and PHC (RQ2)

Dulfer et al. (2015) and Spijkerboer et al. (2007) identified that PCCHD reported comparable or better mental health than normative samples (Dulfer et al.'s study included parents of adolescents with ToF who were well enough to participate in an exercise training

program). Goldbeck and Melches (2005) found that parents of children aged between 7-20, tended to rate their psychological well-being as reasonably high (Mean = 79.2/100, Median 81/100, SD = 11.5). Gronning-Dale and colleagues identified no differences in life satisfaction and well-being between MCCHD and controls during pregnancy and 6 months postpartum (2012), however found that mothers of children with severe CHD had higher levels of anger (2012) and lower subjective well-being (2013; mild-moderate CHD were no different from controls).

Gardner et al. (1996) found that MCCHD (pre-surgery) had significantly higher rates of clinical distress than non-cardiac mothers both at baseline and at a six-month post-operative follow-up. Wray and Sensky (2004) found that PCCHD whose children required surgery were consistently more distressed than parents of healthy controls at pre-surgery. Levels of distress experienced were comparable to those found in parents of children undergoing bone marrow transplant and were consistent regardless of whether the cardiac child was cyanotic or acyanotic. Sklansky et al. (2002) found reduced psychological well-being in pregnant women carrying a foetus with CHD as compared to those who had received normal echocardiography results.

Regardless of timing of diagnosis (pre-/post-natal), Brosig, Whitstone et al. (2007) found that PCCHD were significantly more distressed than normative samples, with prenatally diagnosed parents continuing to be significantly more distressed at a six-month follow up. Pelchat et al. (1999) identified that PCCHD experience more psychological distress than those of healthy children or children with cleft lip/palette but show similar levels of distress to parents of children with Downs Syndrome (DS. Note: 40-60% of DS cases have comorbid CHD and 4-10% of CHD cases also involve DS; Rashid, 2013). Likewise, Arafa, Zaher, El-Dowarty and Moneeb (2008) found the PCCHD have poorer mental health than parents of children with other conditions (HRQoL). Similarly, using the SCL, Lawoko and Soares found that PCCHD

experience levels of distress higher than normative samples (2002); clinically depressed populations (2002); parents of healthy children (2003a); and, for mothers, poorer mental health than those of children with other diseases (2003a). Gardner et al. (1996) found that cardiac mothers showed less positive affect than non-cardiac mothers in video footage of mother-child interactions.

3.2.3 Impact of the timing of diagnosis on psychological distress (RQ3)

In a qualitative study, Cantwell-Bart and Tibballs (2015) found that 83% of parents reported that the time of diagnosis and the "aftermath", were the "worst of their lives" and that they had experienced intense distress during this period. Hearps et al. (2014) found that there was no difference in psychosocial risk for PCCHD based upon pre- versus post-natal diagnosis. Brosig, Whitstone et al. (2007) et al. identified that, whilst PCCHD experienced distress at the time of diagnosis, the timing did not have a significant effect (prenatal - 58% clinically distressed; postnatal 71% clinically distressed). Birth was also identified as a distressing time regardless of time of diagnosis (75% prenatal and 71% postnatal clinically distressed). At six-month follow up, PCCHD with postnatal diagnosis showed a considerable reduction in distress (10% clinically distressed) and had lower levels of distress than prenatal counterparts (45%). Brosig, Whitstone et al. (2007) suggested that prenatal diagnoses are usually associated with more severe heart defects that have greater impact on the child and their parents. Qualitative analysis of clinical interviews indicated themes of anger, disbelief, fear and guilt at diagnosis and birth (regardless of diagnosis timing), along with reflections on the difficulty of parenting within the first six months. Connor et al. (2010) also qualitatively identified distress around uncertainty at the time of diagnosis for PCCHD.

Similarly, Pinto et al. (2016) found psychological distress at diagnosis was higher for PCCHD with a postnatal diagnosis than those with a prenatal diagnosis. Whilst this finding was

consistent for comparisons made at time of birth, it was not apparent at follow up. Fathers of postnatally-diagnosed infants showed significantly higher levels of distress than fathers of prenatally diagnosed infants at diagnosis and birth, however mothers showed no difference at diagnosis. For women, advanced gestational age was associated with higher distress levels.

3.2.4 Impact of CHD severity on psychological distress (RQ4)

Brosig, Whitstone et al. (2007) identified that at time of diagnosis, 81% of individuals whose infant/foetus had severe CHD had clinically significant levels of distress, compared to 33% with less severe CHD. Similarly, Yildiz, Celebioglu and Olgun (2009) and Lawoko and Soares (2002) found that the more severe the CHD, the greater the intensity of distress for PCCHD. Lawoko and Soares (2004) found that increased caregiving burden (typically associated with more severe CHD) was associated with increased psychological distress. In a large Norwegian inception cohort study, Gronning-Dale et al. (2013) found that MCCHD with severe defects had significantly lower self-rated well-being than controls, whereas MCCHD with mild and moderate defects had comparable levels of subjective well-being.

Conversely, Doherty et al. (2009) identified that for PCCHD, diagnostic category and number of symptoms were not predictive of psychological distress. Wray and Sensky (2004) found that cyanotic status was not correlated with distress and Hearps et al. (2014) identified no differences in psychosocial risk for PCCHD based on a single versus biventricular repair. Berant, Mikulincer and Florian (2001) and Berant, Mikulincer and Shaver (2008) found that when CHD was severe, declining mental health over time was predicted by maternal avoidant-attachment at baseline. Goldbeck and Melches (2006) found that whilst CHD severity was not related to quality of life in PCCHD, social disadvantage (which often accompanies CHD) was.

3.2.5 Impact of surgery on psychological distress (RQ5)

Bevilacqua et al. (2013) reported that 60-82% of parents whose infants had undergone cardiac surgery before four months experienced psychological distress, with Wray and Sensky (2004) finding psychological distress in 48-65% of PCCHD prior to surgery. Utens et al. (2002) identified that parents of CHD children undergoing cardiac surgery, showed significantly more psychological distress than parents of CHD children undergoing cardiac catheterisation (less invasive). This difference was consistent for mothers, fathers and parents, and was also present at pre- and post-surgery. Landolt (2011) examined psychological distress at discharge from hospital following open heart surgery and found that PCCHD had reduced mental health as compared to normative samples.

Four studies investigated changes in psychological distress pre-to-post and consistently found a reduction in distress for PCCHD. Utens et al. (2002) found, regardless of gender or type of cardiac intervention (catheterisation versus surgery), psychological distress in PCCHD dropped pre-to-post surgery for mothers, fathers and parents overall. Similarly, Gardner et al. (1996) identified decreases in clinically significant distress pre-surgery (75%) to follow-up (0%) for mothers. Similarly, Wray and Sensky (2004) found that psychological distress decreased significantly pre-to-post surgery for PCCHD (in general and also for the acyanotic sub-group). Landolt et al. (2011) compared psychological distress in PCCHD at post-surgical discharge and six months later, finding that distress had normalised for PCCHD (except for when the child's illness was rated as having a greater impact on the family functioning). In addition, Doherty et al (2009) found that surgical history was not a longer-term predictor of distress in PCCHD.

3.2.6 Influence of parent gender on psychological distress (RQ6)

A number of studies examined gender differences in psychological distress and found mothers consistently more distressed than fathers across time. Bevilacqua et al. (2013) found

81.8% of mothers (versus 60.6% of fathers) experienced clinical levels of psychological distress and Wray and Sensky (2004) found 63% of mothers (versus 48% of fathers) were distressed pre-surgery (25% and 17% post-surgery respectively). Yildiz et al. (2009) and Lawoko and Soares (2002, 2003a) found that mothers experience significantly more distress than fathers. Similarly, Doherty et al. (2009) identified that mothers showed significantly higher rates of clinical distress than fathers (33% versus 18%). Whilst Pinto et al. (2016) found that women carrying a foetus with CHD were more distressed than fathers at time of diagnosis, gender differences were not apparent in those diagnosed postnatally.

3.2.7 Interventions and psychological distress (RQ10)

McCusker et al. (2012) found that mothers who had undertaken the CHIP-school intervention showed less personal strain, emotional problems and psychological distress than controls at a ten-month follow-up.

3.3 Stress (RQ1)

Stress (as a discrete component of psychological distress) was examined by twenty-seven studies (qualitative n=10; quantitative n=12, mixed design n=5. Table 3.3). Twenty-three studies identified stressors for PCCHD, including: perception of reduced child quality of life (Majnemar et al., 2006); caregiving burden (Bruce, Lilja & Sundin, 2014; Pelchat et al., 1999; Pridham et al., 2010); child characteristics that make parenting difficult (Brosig, Mussatto, Kuhn & Tweddell, 2007; Torowicz, Irving, Hanlon, Sumpter & Mendoff-Cooper, 2010); perceptions of parenting competence (Carey et al., 2002; Torowitz et al., 2010); challenges with discipline (Carey et al., 2002; Lee & Rempel, 2011; Majnemar et al., 2006;); reduced social support (Lee, Yoo & Yoo, 2007; Tak & McCubbin, 2002; Werner, Latal, Buechel, Beck & Landolt, 2014); financial pressure (Bruce et al., 2014; Connor et al., 2010; Franck, McQuillan, Wray, Grocott & Goldman, 2010; Sarajuuri, Lonnqvist, Schmidtt, Almqvist & Jokinen, 2012); family member

burden (Brosig, Mussato et al., 2007; Brosig, Whitstone et al., 2007; Bruce & Sundin, 2012; Bruce et al., 2014; Connor et al., 2010; Lan et al.,, 2007; Pridham et al., 2010); gender roles (Clark & Miles, 1999); and diagnosis/medical intervention (Connor et al., 2010; Cantwell-Bartl & Tibballs, 2015; Hoehn et al., 2004; Diffin, Spence, Naranian, Badawi & Johnston, 2016; Franck et al., 2010).

Table 3.3

Studies Addressing Stress in PCCHD

Author (date)	Type	Topic	RQ
Brosig, Mussato et al. (2007)	Quan	Stressors, severity	1, 4
Brosig, Whitstone et al. (2007)	Mixed design	Stressors, COD/HC comparison	1, 2
Bruce & Sundin (2012)	Qual	Stressors	1
Bruce et al. (2014)	Qual	Stressors	1
Bruce et al. (2016)	Qual	Stressors	1
Cantwell-Bartl & Tibballs (2015)	Qual	Stressors, surgery	1, 5
Carey et al (2002)	Mixed design	Stressors, COD/HC comparison	1, 2
Clark & Miles (1999)	Qual	Stressors, surgery	1, 5
Connor et al. (2002)	Qual	Stressors, severity, surgery	1, 4, 5
Diffin et al. (2016)	Quan	Stressors, surgery	1, 5
Dulfer et al. (2015)	Quan	COD/HC comparison	2
Franck et al. (2010)	Mixed design	Stressors, prevalence, COD/HC comparison, surgery, parent gender	1, 2, 5, 6
Hoehn et al. (2004)	Mixed design	Stressors, surgery	1, 5
Kosta et al. (2015)	Qual	Stressors	1
Lan et al. (2007)	Qual	Stressors, surgery	1, 5
Lee & Rempel (2011)	Qual	Stressors	1
Majnemer et al. (2006)	Quan	Stressors, prevalence, severity	1, 4
McCusker et al. (2012)	Quan	Interventions	10
Odegard et al. (2002)	Mixed design	Interventions	10

Pelchat et al. (1999)	Quan	Stressors, COD/HC comparison, severity, parent gender	1, 4, 6
Pridham et al. (2010)	Qual	Stressors	1
Sarajuuri et al. (2012)	Quan	Stressors, COD/HC comparison, severity, parent gender	1, 2, 4, 6
Tak & McCubbin (2002)	Quan	Stressors, severity	1, 4
Torowitz et al. (2010)	Quan	Stressors, COD/HC comparison, severity	1, 2, 4
Uzark & Jones (2003)	Quan	Stressors, COD/HC comparison, severity	1, 2, 4
Vrijmoet-Wiersma et al. (2009)	Quan	COD/HC comparison, severity	2, 4
Werner et al. (2014)	Quan	Stressors, severity, parent gender	1, 4, 6

Majnemar et al. (2006) found that when parents perceive that a child has reduced psychosocial wellbeing, parents have increased levels of stress. Qualitatively, Lee and Rempel (2011) reported that parents found disciplining a CHD child difficult and this related to recognition of the difficulties experienced by the child and wanting to make the child's life as easy as possible. Additionally, Majnemar et al. (2006) found that child problem-behaviours were correlated with stress in PCCHD and Carey et al. (2002) noted that PCCHD experience considerable strain in maintaining energy for parenting and report that their discipline is inadequate. Torowicz et al. (2010) and Brosig, Mussato et al. (2007) identified that children/infants with SV physiology (e.g. HLHS) present with characteristics (such as reduced distractability, mood and adaptability) that made them difficult to parent, and parents of SV children experience levels of stress higher than BV counterparts (Brosig, Mussato et al., 2007; Torowicz et al., 2010). However, whilst Torowicz et al. (2010) and Brosig, Mussato et al. (2007) found that parents of SV children had higher levels of stress relating to parenting competence than BV parents, only Torowicz et al. found that this was higher than controls.

Four studies examined role restriction in relation to stress. Stress related to role restriction was reported Bruce et al. (2014), Pelchat et al. (1999) and Pridham et al. (2010). However,

Brosig, Mussato et al. (2007) found lower levels of role restriction stress in PCCHD than normative samples.

Three studies identified that access to social support can have a positive impact on stress in PCCHD. Werner et al. (2014) and Tak and McCubbin (2002) found that reduced social support was related to increased family stress as rated by PCCHD, and Lee et al. (2007). identified lower stress with increased social connection (internet) and support (in person). Four studies found that factors relating to financial pressures can impact on stress. Bruce et al. (2014) found that PCCHD were fatigued and stressed by financial pressures. Connor et al. (2010) identified higher levels of stress with lower socio-economic-status and Franck et al. (2010) found that stress was highest for PCCHD who were migrants and/or living in financially deprived communities (in the United Kingdom). Sarajuuri et al. (2012) found a correlation between maternal occupation and stress. However, Uzark and Jones (2003) identified that family SES was not a predictor of stress in PCCHD.

Seven studies looked at the impact of CHD on the broader family. Three studies identified burden to the broader family as a source of stress for parents, especially where CHD was complex (Brosig, Mussato et al., 2007; Brosig, Whitstone et al., 2007; Connor et al., 2010). Six studies reported on the 'balancing act' of developing and sustaining family identity/structure/values that encompassed both cardiac and non-cardiac members, whilst also dealing with the responsibilities of the CHD child (Bruce & Sundin, 2012; Bruce et al., 2014; Bruce, Lilja & Sundin, 2016; Kosta et al., 2015; Lan et al., 2007; Pridham et al., 2010).

3.3.1 Prevalence of stress (RQ1)

Majnemar et al (2006) found that 46% PCCHD experience stress, with approximately 25% indicating high levels of stress. Franck et al. (2010), found that PCCHD whose children

were undergoing surgery experienced moderate to high stress consistently across time, with moderately high scores pre-surgery that decreased slightly over time but increased if the child was hospitalised for over fifteen days.

3.3.2 Comparison of stress in PCCHD to PCOD and PHC (RQ2)

Four studies found that PCCHD have significantly higher levels of stress than normative samples of control groups. Torowicz et al. (2010) found that the demands of caring for a CHD child was a significant source of stress for PCCHD compared to that of healthy controls. Sarajuri et al. (2012) also found that PCCHD reported higher levels of total stress than controls, however at a diagnostic level only parents of children with HLHS (severe CHD) differed significantly from the control group. Pelchat et al. (1999) found PCCHD have comparable levels of stress those of children with DS (who can often have CHD as a complication) and significantly higher levels of stress than PHC or those with a cleft-lip/palette (a less severe congenital defect). Franck et al. (2010) found that there were no differences in stress levels for PCCHD undergoing surgery based upon whether their child had DS or not, or on whether there were other congenital abnormalities present. Similarly, Uzark and Jones (2003) identified higher total stress in PCCHD than normative samples, with 17.5% at clinically significant levels.

Four studies identified that overall stress/strain was comparable to (Carey et al., 2002; Vrijmoet-Wiersma, Ottenkamp, van Roozendaal, Grootenhuis & Koopman, 2009), or lower than (Brosig, Whitstone et al., 2007; Dulfer et al., 2015), normative samples and/or healthy controls.

3.3.3 Impact of the timing of diagnosis on stress (RQ3)

No studies specifically examined stress (as a discrete component of psychological stress) in relation to timing of diagnosis.

3.3.4 Impact of CHD severity on stress (RQ4)

One qualitative (Connor et al, 2010) and fourteen quantitative studies provided information regarding the impact of CHD severity on stress in PCCHD. Of these, four focused on SVP such as HLHS (Brosig, Mussato et al., 2007; Sarajuuri et al., 2012; Torowicz et al., 2010; Vrijomoet-Wiersma et al.,2009).

Pelchat et al. (1999) and Tak and McCubbin (2002) found no relationship between level of stress and severity of diagnosis for either mothers or fathers. Similarly, Uzark and Jones (2003) found that parenting stress was not related to the severity of the heart defect, whether the defect was repairable, nor time since last surgery. Vrijmoet-Wiersma et al. (2009) identified that whilst CHD severity was not a predictor of stress in parents, that parents of children with HLHS (severe CHD) had higher ratings than those of children with less severe defects. Similarly, Brosig, Mussato et al. (2007) reported that parents of children with HLHS had greater overall stress than those of children with TGA, and Sarajuuri et al. (2012) found significantly higher stress in parents of children with HLHS than those with functioning UVH. Torowitz et al (2010). found that parents of children with SVP (a more severe form of CHD) were significantly more stressed than those with BVP and control groups.

Qualitatively, Connor et al. (2010) found higher levels of parental stress associated with greater disease complexity and also identified that at 6-7 months post-surgery, parents continued to describe stress as acute and were waiting for the situation to “settle down”. Four quantitative studies also identified medical complications that increased stress, such as seizures (Sarajuuri et al., 2012); preoperative cyanosis (linked to high postoperative stress by a factor of 4.5; Majnemar et al., 2006); and number of medications (Werner et al., 2014). Werner et al (2014) also found that 45% of maternally rated family stress variance was attributable to the presence of the CHD, lower levels of social support and longer hospital stays, and 30% of paternal rated variance was attributable to CHD and lower levels of social support.

3.3.5 Impact of surgery on stress (RQ5)

Seven studies reported on the impact of surgery for PCCHD. The preparation of the child and family for surgery, as well as the undertaking of surgery, was reported as emotionally draining (Connor et al., 2010) and a period of intense stress by three studies (Cantwell-Bartl & Tibballs, 2015; Connor et al., 2010; Lan et al., 2007), with the hospital stay itself considered a period of increased stress (Clark & Miles, 1999). This stress was seen to intensify for PCCHD during the surgery decision-making process (Cantwell-Bartl & Tibballs, 2015) and when multiple surgeries were required (Connor et al., 2010). Within intensive care settings, the sights and sounds of NICU, along with the appearance/behaviour of the child (Diffin et al., 2016; Franck et al., 2010) and parental role (Franck et al., 2010) were seen as the most stressful. Hoehn et al. (2004) also identified that for infants diagnosed with severe CHD at birth (requiring surgery), FCCHD experienced stress relating to the infant and mother being in separate hospitals. Clark and Miles (1999) identified that fathers struggled with gender-role expectations to stay strong for their family, and felt they needed to hide intense emotions ("have to be strong", "men can be weak but no-one realises it…women act confident so I have to be").

3.3.6 Influence of parent gender on stress (RQ6)

Four studies examined stress levels based on the gender of the parent, with mixed results. Franck et al. (2010) found few gender differences over time. Whilst Werner et al. (2014) found no gender differences on overall measures of stress, there were differences in specific domains (fathers reported having less 'ups and downs', less time with family/friends and having 'given up' more aspects of their life than mothers). Similarly, Sarajuuri et al (2012) identified that mothers experienced less child-related, and more parent-related, stress than fathers. Finally, Pelchat et al. (1999) found that mothers consistently experienced more stress than fathers.

3.3.7 Interventions and stress (RQ10)

Two studies reported on the efficacy of interventions designed to reduce stress for PCCHD. Odegard, Modest and Laussen (2002) identified that 96.3% of parents present at their child's anaesthetisation felt it to be a positive experience and important for the reduction of pre-surgery stress. McCusker et al (2012) found that mothers who had undertaken the CHIP-school intervention showed less personal strain than controls at a ten-month follow-up

3.4 Depression (RQ1)

The experience of depression was examined in 18 quantitative studies and one mixed methods design study, with an additional two qualitative studies describing themes relating to symptoms of depression (Table 3.4). Qualitatively, Bruce et al. (2016) identified that fathers of infants with CHD reported feelings of loneliness that were exacerbated by being unable to be directly involved in their child's care (such as when an infant is in NICU) and/or when they perceived that they were lacking in support from other people. Clark and Miles (1999) identified themes of sadness for fathers in relation to their child's diagnosis ("deep hurt inside", "crying" and "sadness").

Table 3.4

Studies Exploring Depression in PCCHD

Author (date)	Type	Topic	RQs
Bruce et al. (2016)	Qual	General themes	1
Clark & Miles (1999)	Qual	General themes	1
Bevilacqua et al. (2013)	Quan	Prevalence, COD/HC comparison, diagnosis, parent gender	1,2, 3, 6
Brosig, Mussato et al. (2007)	Quan	COD/HC comparison, severity	2, 4
Diffin et al. (2016)	Quan	COD/HC comparison, surgery	2, 5
Dulfer et al. (2015)	Quan	COD/HC comparison, surgery	2, 5
Dulfer et al. (2015)	Quan	COD/HC comparison, surgery	2, 5

Jordan et al. (2014)	Mixed design	Surgery	5
Lawoko & Soares (2002)	Quan	Prevalence, COD/HC comparison, severity, parent gender	1, 2, 4, 6
Lawoko & Soares (2003a)	Quan	Severity	4
Lawoko & Soares (2003b)	Quan	Severity	4
Lawoko & Soares (2004)	Quan	Severity	4
Lawoko & Soares (2006)	Quan	Severity, prevalence	1, 4
Pinto et al. (2016)	Quan	Diagnosis, parent gender	3, 6
Rona et al. (1998)	Quan	Prevalence, COD/HC comparison, diagnosis,	1, 2, 3
Rychik et al. (2013)	Quan	Prevalence	1
Solberg et al. (2011a)	Quan	Prevalence COD/HC comparison, severity	1, 2 , 4
Solberg et al. (2011b)	Quan	COD/HC comparison, severity, surgery	2, 4, 5
Solberg et al. (2012)	Quan	COD/HC comparison, severity, surgery	2, 4, 5
Spijkerboer et al. (2007)	Quan	COD/HC comparison	2
Utens et al. (2002)	Quan	COD/HC comparison, parent gender, surgery	2, 5, 6
Yildiz et al. (2009)	Quan	Severity, parent gender	5, 6

3.4.1 Prevalence of depression (RQ1)

Lawoko and Soares (2002) found that 18% of PCCHD had scores for depression within or above the rates for a psychiatric outpatient population. Further, Lawoko and Soares (2006) found that 13% of PCCHD had clinical levels of depression that had persisted for at least 12 months, with 7% having longstanding hopelessness at levels seen in depressed populations. The prevalence rate for hopelessness in PCCHD ranged from 11-16%, depending on the timing of the assessment and the demographic of the sub-group.

Five studies identified 18-45.7% of women with a CHD foetus/child as having clinical levels of depression. Rychick et al. (2013) found that 22% of women carrying a foetus diagnosed with CHD within the previous 2-4 weeks were clinically depressed, whereas Bevilacqua et al. (2013) found that the rate for women with a recently diagnosed infant or foetus was 45.7%.

Solberg et al. (2011a) found that 29.5% of mothers of infants with severe CHD had PND, with Rona, Smeeton, Beech, Barnett & Sharland (1998) finding 18% of mothers of CHD infants were clinically depressed. Further, Lawoko and Soares (2002) identified that 21% of mothers of CHD infants were depressed at levels comparable to or above those of psychiatric outpatients.

3.4.2 Comparison of depression in PCCHD to PCOD and PHC (RQ2)

Eleven studies examined depression in PCCHD as compared to PHC or PCOD. Lawoko and Soares (2002), in a study of children with varied illness severity and requirements for surgery, found significantly higher levels of depressed symptomology (and rates of clinical depression) in PCCHD than in PHC, PCOD and clinically depressed people and psychiatric outpatients. Diffin et al. (2016) found higher rates of overall, moderate and severe depression in PCCHD as compared to health controls across time (in NICU at time of surgery, and six and twelve-month follow-up) compared to healthy controls. Rona et al. (1998) found depression in 18% of MCCHD, a rate higher than women carrying a foetus with CHD and those with a false positive CHD screen. Bevilaqua et al. (2013) identified that pregnant women with a foetus recently diagnosed with CHD (2-4 weeks prior) reported significantly higher levels of depression than normative samples. In a longitudinal study, Solberg et al. (2011a, 2011b, 2012) found mothers of infants with severe CHD were consistently and significantly more depressed than those of the healthy cohort across time. However, these differences were not apparent between MHC and mothers of children with mild-moderate CHD across time (prenatally and six, eighteen and thirty-six months postpartum).

Similarly, Dulfer et al. (2015) found no significant difference in levels of severe depression between PCCHD (of adolescents engaging in an exercise intervention program) and a normative sample, with Utens et al. (2002) finding significantly lower levels of serious depression in PCCHD than in normative samples and Brosig, Mussato et al. (2007) also lower

levels of depression in PCCHD. It should be noted, however, that in these examples, children had already undergone successful cardiac surgery/catheterisation (Utens et al., 2002; Brosig, Mussato et al., 2007; Dulfer et al., 2015). Nevertheless, Spijkerboer et al. (2007) also found lower levels of clinically significant depression in PCCHD than in a reference group.

3.4.3 Impact of the timing of diagnosis on depression (RQ3)

Three studies examined the impact of prenatal versus postnatal diagnosis on ratings of depression in PCCHD, with inconsistent results. Bevilacqua et al. (2013) found no differences in depression scores based upon timing of diagnosis. Similarly, Pinto et al. (2016), also found no significant differences between groups across time (diagnosis, birth and six-month follow-up), however noted a peak at birth and a decline in depression at six-month follow-up for both groups. Conversely, Rona et al. (1998) found that clinical levels of depression were highest among mothers of postnatally diagnosed children with severe malformations of the heart (18%), than in those diagnosed prenatally (4%) or in those given a false-positive prenatal diagnosis (5%).

3.4.4 Impact of CHD severity on depression (RQ4)

Nine studies examined the relationship of CHD severity to the experience of depression in PCCHD. Of these, seven reported on two separate study clusters: a longitudinal study of Norwegian mothers of children with CHD (Solberg et al., 2011a, 2011b, 2012) and an exploration of various predictors of psychological distress factors in Swedish PCCCHD (Lawoko & Soares, 2002; 2003b; 2004; 2006). In general, it was indicated that the more severe the CHD, the more likely it is that PCCCHD will experience symptoms of depression but that the relationship may not be a directly causal one.

Brosig, Mussato et al. (2007) found that parents of children with HLHS had significantly higher levels of depression than those whose children had TGA, and Yildiz et al (2009) reported

that severe defects were related to significantly greater depression than mild-moderate defects. Further, in a longitudinal study by Solberg and colleagues, mothers of infants with severe CHD had more clinically significant symptoms of PND than mothers of infants with mild and moderate CHD (2011a) and showed consistent elevations of depression at six, eighteen and thirty-six-month follow-up that were not found in the overall cohort, nor identified in the mild to moderate severity groups (2011b, 2012).

However, Lawoko and Soares (2006) indicated that a causal relationship between CHD severity and symptoms of depression is more difficult to identify. More specifically, regression analysis indicated that depression (at a single point in time) was influenced by the severity of diagnosis (Lawoko & Soares, 2002), however longstanding morbidity was not similarly explained (Lawoko & Soares, 2006). Factors such as caregiving burden and financial instability (likely to be more prevalent with severe CHD) were better predictors of long-term symptoms than severity of diagnosis itself, as were dissatisfaction with care and social isolation (Lawoko & Soares, 2004; 2006). Limitations in the access to social supports shared a reciprocal relationship with increased depression and hopelessness in PCCHD, however was not in itself directly predicted by disease severity (Lawoko & Soares, 2003b; 2006). Lawoko & Soares (2006) suggested that the influence of factors such as caregiving burden might provide an explanation for this.

3.4.5 Impact of surgery on depression (RQ5)

The relationship between surgery and symptoms of depression was examined in seven studies with mixed findings. Utens et al. (2002) found that PCCHD undergoing cardiac catherization (less invasive proceedure) were less depressed than PCCHD undergoing cardiac surgery (more invasive) when measured pre- and post-proceedure, however both groups were

consistently lower than normative samples across time. Dulfer et al. (2015) found no significant differences in severe depression between post-surgery PCCHD and normative samples.

In contrast, longitudinal studies by Diffin et al. (2016) and Solberg et al (2011b; 2012) found differences between in levels of parental depression between PCCHD and normative/control groups. Specifically, Diffin et al. (2016) found that PCCHD whose infant was admitted to NICU for surgery, had significantly higher percentages of moderate and severe depression than non-cardiac controls and that, whilst this difference was not apparent at six-month post-surgery follow up, it was once again apparent at twelve months. Solberg et al. (2011b, 2012) found that mothers of infants that had undergone surgery for severe CHD, consistently reported greater depression than the general population over time (6, 18 and 36 months). Similarly, Utens et al (2002) reported that MCCHD undergoing cardiac surgery did not show the decrease in depression pre- to post-surgery identified in MCCHD undergoing catherization. Jordan et al. (2014) found that MCCHD who had had surgery before three months of age were more likely to experience PND if they experienced reduced infant attachment.

3.4.6 Influence of parent gender on depression (RQ6)

Five studies examined gender differences and found higher rates of depression in mothers than fathers (Bevilacqua et al., 2013; Lawoko & Soares, 2002; Pinto et al., 2016; Utens et al., 2002; Yildiz et al., 2009). For example, Bevilacqua et al. (2013) found significantly higher clinical depression in mothers (45.7%) than fathers (20%). Lawoko and Soares (2006) reported that mothers more likely to be depressed over time than fathers. Similarly, Pinto et al. identified that mothers whose children was diagnosed prenatally were more depressed over time than fathers.

3.4.7 Interventions and depression (RQ10)

No studies evaluated interventions for depression in PCCHD.

3.5 Anxiety (RQ1)

The experience of anxiety in PCCHD was examined in 28 studies (quantitative n=15; qualitative n=9; mixed design n=4, Table 3.5). Qualitative studies identified themes of constant worry and concern (Bruce et al., 2014; Carey et al., 2002; Clark & Miles, 1999; Lan et al., 2007; Pinelli, 1981; Pridham et al., 2010; Rempel & Harrison, 2007; Rempel, Harrison & Williamson, 2009; Sparacino et al., 1997); uncertainty (Carey et al., 2002; Clark & Miles, 1999; Rempel et al., 2009; Rempel & Harrison, 2007; Sparacino et al., 1997); fear and terror (Clark & Miles, 1999; Sparacino et al., 1997) and vulnerability (Bruce et al., 2014; Clark & Miles, 1999). Bright et al. (2013; mixed design) identified that 17% of FCCHD experience apprehension and condition-specific worry regarding their infant.

Table 3.5

Studies Exploring Anxiety in PCCHD

Author (date)	Type	Topic	RQ
Bright et al (2013)	Mixed design	General themes	1
Bruce et al. (2014)	Qual	General themes	1
Carey et al (2002)	Mixed design	General themes	1
Clark & Miles (1999)	Qual	General themes	1
Diffin et al. (2016)	Quan	COD/HC comparison, surgery	2, 5
Fischer et al. (2012)	Quan	Prevalence	1
Hoehn et al. (2004)	Mixed design	Diagnosis	3
Lan et al. (2007)	Qual	General themes	1
Lawoko & Soares (2002)	Quan	COD/HC comparison, parent gender	2, 6
Lawoko & Soares (2006)	Quan	Prevalence, parent gender	1, 6
Lok & Menahem (2004)	Qual	Diagnosis	3
McCrossan et al (2008)	Mixed design	Interventions	10

McCusker et al. (2010)	Quan	Prevalence, interventions	1, 10
Pinelli (1981)	Qual	General themes	1
Pinto et al. (2016)	Quan	Diagnosis, severity, surgery, parent gender	3, 4, 5, 6
Pridham et al. (2010)	Qual	General themes	1
Rempel & Harrison. (2007)	Qual	General themes	1
Rempel et al. (2009)	Qual	General themes	1
Rahimianafar et al. (2015)	Quan	COD/HC comparison, surgery	2, 5
Rona et al. (1998)	Quan	COD/HC comparison	2
Rychik et al. (2013)	Quan	Prevalence	1
Sklansky et al. (2002)	Quan	COD/HC comparison	2
Solberg et al. (2011b)	Quan	COD/HC comparison	2
Solberg et al. (2012)	Quan	COD/HC comparison	2
Sparacino et al. (1997)	Qual	General themes	1
Spijkerboer et al. (2007)	Quan	COD/HC comparison	2
Utens et al. (2002)	Quan	Surgery, parent gender	5, 6
Vrijmoet-Wiersma et al. (2009)	Quan	COD/HC comparison, severity, surgery, parent gender	2, 4, 5, 6

3.5.1 Prevalence of anxiety (RQ1)

Four studies found rates of clinically significant anxiety in PCCHD that ranged between 5-31% (Fischer et al., 2012; Lawoko & Soares, 2006; McCusker et al., 2010; Rychik et al., 2013). Lawoko and Soares (2006) found that 7% of PCCHD had clinical levels of anxiety (SCL-90R) that had persisted for at least 12 months, with prevalence rates ranging between 11-18% depending on gender and timing of the assessment. McCusker et al. (2010) found rates of state anxiety (STAI) ranged between 20-22% over a six-month period among parents not receiving interventions, and Rychik et al. (2013) reported state anxiety in 31% of MCCHD. Conversely, Fischer et al. (2012) found clinically significant state anxiety in 5% of PCCHD (with an additional 1% within the ‘borderline’ range).

3.5.2 Comparison of anxiety in PCCHD to PCOD and PHC (RQ2)

Ten studies compared anxiety in PCCHD to PHC/PCOD and generally found that PCCHD experienced higher levels of anxiety. Lawoko and Soares (2002) found that PCCHD experience significantly more anxiety than PHC and have significantly higher rates of clinical anxiety (15%) than PHC (7%) and PCOD (9%), with findings also consistent across genders. Sklansky et al. (2002) identified higher anxiety in women with a prenatal diagnosis of CHD than those who had received normal echocardiography. Diffin et al. (2016) found that over a twelve-month period, PCCHD had greater anxiety than a control group, with moderate-severe anxiety significantly higher at twelve-month follow-up. Rona et al (1998) found that 62% of women with a CHD infant/foetus showed significant anxiety, a rate higher than a false-positive group (30%). Solberg et al. (2011b; 2012) also found significant elevations in anxiety among PCCHD when compared to the overall cohort at six, eighteen and thirty-six-month follow-up. Rona et al. (1998) found that anxiety was higher in mothers of CHD infants and in women carrying a CHD foetus, than in those with a false positive CHD screen, and Rahimianafar et al. (2015) reported that levels of anxiety were higher than normative samples in PCCHD. Vrijmoet-Wiersma et al. (2009) found that state anxiety was higher for MCCHD than reference groups, however was similar for FCCHD. In contrast, Dulfer et al. (2015) and Spijkerboer et al. (2007) found that PCCHD were less anxious than reference groups.

3.5.3 Impact of timing of diagnosis on anxiety (RQ3)

Three studies examined the role of prenatal versus postnatal diagnosis in parental anxiety for a child with CHD. Qualitatively, Lok and Menahem. (2003) identified diagnosis as a time of heightened anxiety for PCCHD. Hoehn et al. (2004) found that at time of infant surgery, MCCHD diagnosed prenatally and postnatally showed no difference in anxiety, however prenatally diagnosed FCCHD had lower anxiety than those diagnosed postnatally. Pinto et al.

(2016) identified that the older the gestational age for a foetus prenatally diagnosed with CHD, the more anxious the parent; however, PCCHD of prenatally diagnosed infants were less anxious at diagnosis and birth (but not a follow-up) than those diagnosed postnatally (significant only for fathers)

3.5.4 Impact of CHD severity on anxiety(RQ4)

Two studies examined CHD severity in relation to anxiety in PCCHD and found that it was not a risk factor (Pinto et al., 2016; Vrijmoet-Wiersma et al., 2009).

3.5.5 Impact of surgery on anxiety (RQ5)

Five studies examined the anxiety in PCCHD in relation to surgery/hospitalization. Utens et al. (2002) identified that parents of children who required more invasive interventions (cardiac surgery) were consistently more anxious across time than those who required less invasive interventions (cardiac catheterisation). However, regardless of level of invasiveness, anxiety levels reduced over time for MCCHD following the cardiac procedure. Consistent with this, Pinto et al. (2016) found that anxiety for PCCHD decreased over time for PCCHD regardless of severity/interventions. Similarly, Vrijmoet-Wiersma et al. (2009) found that for parents of children with complex CHD, time since the last surgical procedure, along with the number of procedures, was a risk factor for anxiety (such that multiple procedures in close proximity to the current time increased anxiety). In contrast, Rahimianafar et al. (2015) identified that anxiety did not significantly differ based on infant hospitalisation history for MCCHD, with Diffin et al. (2016) finding that level of anxiety was correlated more with concerns regarding infant behaviour and appearance in NICU.

3.5.6 Influence of parent gender on anxiety (RQ6)

Five studies looked at the role of parent gender and anxiety and consistently found that mothers experience greater levels of anxiety than fathers. Lawoko and Soares (2002) found that

the percentage of mothers (18%) having levels of anxiety above/within levels for psychiatric outpatients was greater than that for fathers (11%). This persisted across time with significantly more mothers (10%) than fathers (3%) having longstanding clinical anxiety (Lawoko & Soares, 2006). Utens et al. (2002) found that regardless of invasiveness of intervention (cardiac surgery versus catherization), mothers are more anxious than fathers. Pinto et al. (2016) identified that when diagnosis is made prenatally, mothers are more anxious than fathers, and Hoehn et al. (2004) found that prenatal diagnosis is related to more anxiety among mothers at time of infant surgery than fathers. Vrijmoet-Wiersma et al. (2009) identified that state anxiety scores are significantly higher than those found in normative populations for mothers but not for fathers of children with CHD.

3.5.8 Interventions and anxiety (RQ10)

Two studies investigated the effectiveness of interventions designed to reduce parental anxiety. McCusker et al. (2010) found that clinical levels of anxiety in PCCHD reduced significantly (from 26% to 3%) following the CHIP intervention (narrative therapy, parent skills training and psychoeducation) but showed less reduction in the control group (30% to 22%). McCrossan et al. (2008) found that video-conferenced medical consultations (that provided timely advice to parents who were concerned about their child) resulted in a mean decrease in anxiety pre-to-post session.

3.6 Somatisation

Nine quantitative studies examined symptoms of somatisation in PCCCD (Table 3.6). A number of risk factors for somatisation were identified, including CHD severity (fathers. Yildiz et al., 2009); invasiveness of proceedure (fathers. Utens et al., 2002), receipt of benefits (Lawoko & Soares, 2002); financial concerns (Lawoko & Soares, 2002; Yildiz et al., 2009); time (Lawoko & Soares, 2002); lower social availability and interaction (Lawoko & Soares, 2003b); and being

a young parent (fathers aged 20-29 years. Yildiz et al., 2009). Age of the child was not found to be a risk factor for somatisation by Utens et al. (2002) or Yildiz et al. (2009).

Table 3.6

Studies Exploring PCCHD Somatisation

Author (date)	Type	Topic	RQ
Arafa et al. (2008)	Quan	Prevalence	1
Dulfer et al. (2015)	Quan	Prevalence	1
Lawoko & Soares (2002)	Quan	Risk factors	1
Lawoko & Soares (2003b)	Quan	Risk factors	1
Lawoko & Soares (2006)	Quan	Prevalence, parent gender	1, 6
Pinto et al. (2016)	Quan	Diagnosis, parent gender	3, 6
Spijkerboer et al. (2007)	Quan	Prevalence, parent gender	1, 6
Utens et al. (2002)	Quan	Risk factors, surgery	1, 5
Yildiz et al. (2009)	Quan	Risk factors, severity, parent gender	4, 6

3.6.1 Prevalence of somatisation (RQ1)

Lawoko & Soares (2006) identified that between 31-43% of parents (depending on gender and timing of the measure) reported clinical symptoms of somatization, with longstanding symptoms (twelve months or more) reported by 16% to 27% of parents.

3.6.2 Comparison of somatisation in PCCHD to PCOD and PHC (RQ2)

One study examined somatization in PCCHD as compared to PCOD, finding that PCCHD reported higher levels of bodily pain (Arafa et al., 2008). Two studies examined PCCHD in comparison to normative/reference groups and found lower levels in PCCHD (Spijkerboer et al., 2007; Dulfer et al., 2015).

3.6.3 Impact of timing of diagnosis on somatisation (RQ3)

One study looked at the impact of pre- versus post-natal diagnosis on somatization in PCCHD and found that timing of diagnosis was not related to changes in somatization over time (Pinot et al.,2016).

3.6.4 Impact of CHD severity on somatisation (RQ4)

CHD severity was found to be a risk factor for somatisation in FCCHD by Yildiz et al. (2009).

3.6.5 Impact of surgery on somatisation (RQ5)

One study examined the impact of surgery on somatization. Utens et al. (2002) found that the more invasive the surgical procedure for CHD was, the higher the level of somatization in FCCHD. However, whilst ratings of somatisation were higher for children undergoing cardiac surgery versus catheterization, these ratings decreased for the cardiac surgery group following surgery.

3.6.6 Influence of parent gender on somatisation (RQ6)

Four studies examined differences in somatisation based on gender and found that mothers experience higher levels than fathers (Lawoko & Soares, 2006; Pinto et al., 2016; Spikerboer et al., 2007; Yildiz et al., 2009). For example, Lawoko and Soares (2006) found that clinically significant symptoms for mothers increased across a twelve-month period (32% to 43%) and were consistently and significantly higher than those experienced by fathers. Pinto et al. (2016) also reported significantly higher somatisation symptoms at diagnosis, birth and follow-up (4-9 months) for mothers (but only for those diagnosed prenatally).

3.6.8 Interventions and somatisation (RQ10)

No studies examined interventions for somatization.

3.7 Trauma (RQ7)

The experience of 'trauma' was specifically examined in four quantitative studies (Franich-Ray et al., 2013; Helfricht et al., 2009; Landolt, 2011; Rychik et al., 2013) and one qualitative study of vigilance/hypervigilance (Meakins, Ray, Hegadoren, Rogers & Rempel, 2005). A further eleven studies described parental experiences that are consistent with specific trauma symptomology (qualitative n=8; quantitative n=2; mixed design n=1). Together these studies consistently indicate that PCCHD experience trauma related symptomology at clinically significant levels (Table 3.7).

Table 3.7

Studies Related to Trauma in PCCHD

Author (date)	Type	Topic	RQ
Carey et al (2002)	Mixed design	Symptoms	7
Clark & Miles (1999)	Qual	Symptoms	7
Diffin et al. (2016)	Quan	Symptoms	7
Franich-Ray et al. (2012)	Quan	Diagnosis	7
Harvey et al. (2013)	Qual	Symptoms	7
Helfricht et al. (2009)	Quan	Diagnosis	7
Landolt (2011)	Quan	Diagnosis	7
Lee et al. (2007)	Quan	Symptoms	7
Lee & Rempel (2011)	Qual	Symptoms	7
Meakins et al. (2015)	Qual	Symptoms	7
Pridham et al. (2010)	Qual	Symptoms	7
Rempel & Harrison. (2007)	Qual	Symptoms	7
Rychik et al. (2013)	Quan	Diagnosis	7
Sparacino et al. (1997)	Qual	Symptoms	7
White et al. (2016)	Qual	Symptoms	7

3.7.1 Clinically significant trauma (RQ7)

One study examined PTSD in parents whose child had recently been discharged from hospital following open-heart surgery and found that 25.3% of mothers and 25.8% of fathers met the criteria for full PTSD (surgery related), with an additional 32.6% of mothers and 34% of fathers meeting partial requirements for PTSD (measured by the PTDS; Landolt, 2011). Two studies investigated Acute Stress Disorder finding considerable manifestation of the disorder in PCCHD. Specifically, Helfricht et al. (2009) found that 25% of PCCHD met diagnostic threshold for Acute Stress Disorder (using the ASDS), which was significantly more than adult cardiac patients (4%). Franich-Ray et al. (2012) reported that 33.8% of mothers as compared to 18.2% of fathers met diagnostic threshold, with 83% of parents identifying at least one symptom of Acute Stress Disorder at a clinical level and only 11.4% having just one. These findings are similar to Rychik et al. (2013) who reported on clinically important traumatic stress in 39% of MCCHD.

Franich-Ray et al. (2012) found that symptoms of dissociation were the most frequently reported symptoms of Acute Stress Disorder, with each symptom in the dissociation cluster of the ASDS endorsed by at least 26% of parents. In contrast, Rychik et al. (2013) found denial to be the symptom most frequently associated with traumatic stress in MCCHD, as measured by the IES-R.

Helfricht et al. (2009) found that the internal consistency of the avoidance cluster in ASDS was relatively low in PCCHD and suggested that the inescapability of physical illness may account for this. More specifically, the necessity for parents to attend ongoing medical appointments and procedures, and to be actively involved in the medical care of their child, meant that they were less able to engage in physical forms of avoidance (as measured by the ASDS). Helfrict et al. therefore argued that a four-factor model of trauma, which encompasses

symptoms of both physical avoidance (active avoidance) and numbing (passive avoidance) would provide a measure potentially more reflective of the experience of PCCHD.

3.7.2 Evidence of trauma symptomology (RQ7)

A number of qualitative studies described parental experiences that were suggestive of specific trauma symptomology such as hypervigilance and flashbacks. The role of vigilance in parents of children with HLHS was investigated by Meakins et al. (2015) and clear reports of persistent parental hypervigilance were also identified. Themes related to, or parent reports of, hypervigilance were also found in an additional eight qualitative studies (Carey et al., 2002; Clark & Miles, 1999; Lee & Rempel, 2011; Meakins et al., 2015; Pridham et al., 2003; Rempel & Harrison, 2007; Sparacino et al.,1997; White, Moola, Kirsh & Faulkner, 2016). Common themes included hypervigilance based around fear that: the child would be harmed or die and a sense of parental responsibility for the prevention of this (Carey et al., 2002; Clark & Miles, 1999; Lee & Rempel, 2011; Meakins et al., 2015; Rempel & Harrison, 2007; Sparacino et al., 1997); the child was unable to cope (Lee & Rempel, 2011); or alternative carers and/or medical professionals were inept (Carey et al., 2002; Meakins et al., 2015; Rempel & Harrison, 2007; Pridham et al., 2003). White et al. (2016) identified hypervigilance within the context of parents allowing their children to independently attend a camp for children with CHD.

Whilst hypervigilance was identified as originating in an appropriate need for high levels of vigilance by PCCHD (e.g. care of a medically fragile infant), a number of studies identified that parents found it difficult to adjust levels of vigilance over time to match changes in the requirement for vigilance (e.g. after stabilisation of the heart through surgery, or as the child grew older and was able to more independently take care of themselves. Meakins et al., 2015; Sparacino et al., 1997; White et al., 2016); had difficulty judging appropriate levels of vigilance in the face of uncertainty (Carey et al., 2002; Clark & Miles, 1999; Lee & Rempel, 2011;

Meakins et al., 2015; Sparacino et al., 1997; Rempel & Harrison, 2007); or used hypervigilance as a way of attempting to control a situation in which they had very little control (e.g. quarantining the family pre-surgery to prevent surgical delays due to child sickness. Lee & Rempel, 2011; Meakins et al., 2015; Pridham et al., 2010).

Flashbacks were specifically reported by Harvey et al. (2013) in which the 'emotional rollercoaster' of the CHD surgical experiences was noted to have existed in flashbacks for months to years after the event. Whilst not described specifically within the context of a trauma response, Diffin et al. (2016) and Franck et al. (2010) reported that the sights and sounds of NICU, along with the appearance of the infant, were the most distressing aspects of NICU for PCCHD and it is possible that these experiences may have acted as triggers for emotional responses in PCCHD.

3.8 Post-Traumatic Growth (RQ11)

There were no studies that specifically examined Post-Traumatic Growth (PTG) in PCCHD as a construct, nor defined a theme uniquely reflective of the experience of PTG. However, fourteen studies reported on parental responses that could be interpreted as suggestive of the way in which parenting a child with CHD can be transformative in nature (qualitative n=10, quantitative n=2, mixed design n=2. Table 3.8).

Table 3.8

Studies with Themes Related to PTG in PCCHD

Author (date)	Type	Topic	RQ
Brosig, Mussato et al. (2007)	Quan	Themes	8
Bruce et al. (2012)	Qual	Themes	8
Bruce et al. (2014)	Qual	Themes	8
Carey et al (2002)	Mixed design	Themes	8
Clark & Miles (1999)	Qual	Themes	8

Harvey et al. (2013)	Qual	Themes	8
Jordan et al. (2014)	Mixed design	Themes	8
Kocyildirim et al. (2007)	Qual	Themes	8
Lan et al. (2007)	Qual	Themes	8
Lee & Rempel (2011)	Qual	Themes	8
Meakins et al. (2015)	Qual	Themes	8
Rempel & Harrison. (2007)	Qual	Themes	8
Sira et al. (2014)	Quan	Themes	8
Sparacino et al. (1997)	Qual	Themes	8

Three studies specifically reported on the benefits and blessings of having a child with CHD. Brosig, Mussato et al. (2007) found that ninety-six percent of parents identified specific benefits relating to their child's CHD, particularly in feeling closer to their child because of the challenges inherent in the CHD journey. Similarly, Harvey et al. (2013) identified that only through experiencing the "unthinkable", were mothers of CHD children able to find acceptance in the situation and then focus on the benefits and blessings of being a MCCHD. Jordan et al. (2014) reported on themes of increased emotional bonds.

Six studies reported on parental experiences of gratitude and appreciation (including a renewed or newfound understanding of what was considered important in life and/or a sense of celebration of day to day experiences and milestones). These studies encompassed parental attitudes towards their CHD child, family, the supports in their life and the medical professionals involved in the care of their child (Bruce et al., 2012; Carey et al., 2002; Clark & Miles, 1999; Harvey et al., 2013; Kocyildirim, 2007; Lee & Rempel, 2011; Rempel & Harrison, 2007; Rempel et al., 2009; Sira, Desai, Sullivan & Hannon, 2014). Five studies identified a newly developed, re-invigorated or strengthened spiritual connection (expressed through religion, faith and/or connection to God. Harvey et al., 2013; Rempel & Harrison, 2007; Lee & Rempel, 2011;

Sira et al., 2014; Sparacino et al., 1997) and three explored the experience of 'meaning finding' (Lan et al., 2007; Lee & Rempel, 2011; Sparacino et al., 1997). For example, one parent identified "my faith is stronger, not because He [God] allowed our daughter to stay but because I came closer to him" (Harvey et al., 2013).

A sense of 'mastery' or pride in the attainment of complex skills, and the active pursuit of learning through curiosity or the seeking of knowledge was identified by parents in three studies (Bruce et al., 2014; Kocyilidirim et al., 2007; Meakins et al., 2015). 'Paying it forward' (a process where PCCHD experienced satisfaction, pleasure or meaning through the communication of CHD knowledge or shared experience to support others) was found in three (Bruce et al., 2012, 2014; Sira et al., 2014).

3.9 Coping and Resiliency (RQ9)

Twelve quantitative and thirteen qualitative studies explored coping in PCCHD, whilst an additional five studies (qualitative n=3, quantitative n=1, mixed design n=1) examined interventions designed to increase parental well-being (Table 3.9).

Table 3.9

Studies Related to Strategies, Interventions and Barriers for Coping and Resiliency

Author (date)	Type	Coping	RQ
Berant et al. (2001)	Quan	Helpful, unhelpful	9
Berant et al. (2003)	Quan	Unhelpful	9
Bratt et al. (2015)	Qual	Helpful	9
Bruce et al. (2014)	Qual	Helpful	9
Bruce et al. (2016)	Qual	Helpful	9
Clark & Miles (1999)	Qual	Helpful	9
Davis et al. (1998)	Quan	COD/HC comparison	2, 9
Diffin et al. (2016)	Quan	COD/HC comparison	2, 9

Doherty et al. (2009)	Quan	Parent gender, strategies, unhelpful	6, 9
Gudmundsdottir et al. (1996)	Qual	Helpful	9
Harvey et al. (2013)	Qual	Helpful	9
Kocyildirim et al. (2007)	Qual	Interventions	9, 10
Lan et al. (2007)	Qual	Helpful	9
Lee & Rempel (2011)	Qual	Helpful	9
McCrossan et al (2008)	Mixed design	Interventions	9,10
Meakins et al. (2015)	Qual	Helpful	9
Pridham et al. (2010)	Qual	Helpful	9
Redshaw et al. (2011)	Qual	Helpful, interventions	9, 10
Rempel & Harrison. (2007)	Qual	Helpful	9
Rychik et al. (2013)	Quan	Unhelpful	9
Sira et al. (2014)	Quan	Strategies, helpful	9
Sparacino et al. (1997)	Qual	Helpful	9
Spijkerboer et al. (2007)	Quan	COD/HC comparison	2/9
Svavarsdottir & McCubbin (1996)	Quan	Severity, parent gender	4, 6, 9
Tak & McCubbin (2002)	Quan	Severity, helpful	4, 9
Utens et al. (2002)	Quan	Parent gender	6,9
Wilson (2015)	Quan	Interventions	9, 10
Wray & Sensky (2004)	Quan	Unhelpful	9
White et al. (2016)	Qual	Interventions	9,10

3.9.1 Comparison of coping in PCCHD to PHC and PCOD (RQ2 and RQ9)

Two studies compared the coping styles of PCCHD to control/normative groups (Diffin et al., 2016; Spijkerboer et al., 2007), one explored similarities between PCCHD and parents of children with other chronic illness (Davis et al., 1998). Using the UCL, Spijkerboer et al. (2007) reported that PCCHD had more favourable coping mechanisms than a normative sample and showed less tendency to use coping styles such as reassuring thoughts. Whilst Diffin et al. (2016) identified that MCCHD and FCCHD had significantly less avoidance focused coping than a

control group (and that this was consistent across time), they also found that they had lower task focused coping (also consistent across time). Davis et al. (1998) found that a reliance on palliative coping style (emotion-focused, avoidance, wishful thinking and self-blame) along with a perception of high daily stress, reduced maternal psychological functioning and was similar to findings for other chronic illnesses (including Cystic Fibrosis).

3.9.2 CHD severity and coping (RQ4 and RQ9)

Two studies examined CHD severity. Svavarsdottir and McCubbin (1996) identified that the more severe the heart condition, the more likely it was that parents would use a style of coping based upon understanding the healthcare situation. Tak and McCubbin (2002) found that the severity of the CHD was not related to maternal ability to cope.

3.9.3 Influence of parent gender on coping (RQ6 and RQ9)

Doherty et al. (2009) found that mothers used the following coping styles more than fathers: instrumental social support; emotional social support; religion and venting. Alcohol was the least used strategy for mothers and fathers (but was used more by fathers) and there were no differences in use of active, planning, suppression, positive reinterpretation, restraint, acceptance, denial, mental disengagement, behavioural disengagement and humour. Svavarsdottir and McCubbin (1996) found that there were no differences between mothers and fathers in their use of strategies such as ‘strengthening family life and relationships’ and ‘managing psychological tensions’; however, mothers reported more helpful coping behaviours relating to ‘understanding the health care condition’ than fathers. Further, fathers with higher caregiving demands, had higher family systems demands and found it more helpful to use all three coping patterns, a relationship that was not found for mothers.

Svavarsdottir and McCubbin (1996). reported that fathers of younger infants reported higher care-giving demands and also reported more helpful coping strategies related to family,

self and the health care situation. This relationship was not apparent for mothers. Whilst Utens et al. (2002) did not find that age of child when they underwent CHD intervention (surgery or catheterisation) was related to changes in style of coping in their parents, they did identify that PCCHD showed improvements in a palliative style of coping pre-to-post surgery and that fathers (but not mothers) showed an increased use of reassuring thoughts.

3.9.4 Coping strategies used (RQ9)

Doherty et al. (2009) identified alcohol as the least frequent coping strategy used by MCCHD and FCCHD. Sira et al. (2014) found that 1.1% of PCCHD used a coping style defined as ‘maintaining family integration, cooperation and optimism’; 23.4%% used a style defined as ‘maintaining own self-esteem and psychological stability’; and 83.4% used a coping pattern defined as ‘medical communication and consultation efforts’. In addition, 39% indicated that spirituality was an important coping mechanism, as compared to 25.1% who did not find it helpful. Further, 69.7% of mothers used the internet as a source for seeking medical information and 17.3% proactively used online support forums. PCCHD with high levels of coping (based upon family integration, optimism and cooperation), also had a higher reliance on spirituality and reported a stronger sense of coping that came from psychological stability, self-esteem and social support. Likewise, those who emphasised the importance of consultation to understand the medical situation, had more use of the internet and more effective family cooperation and integration. In addition, 27.7% MMCHD used a support group and 4% who did not have one available indicated that they would utilise one if it was.

3.9.5 Helpful coping strategies (RQ9)

Berant et al. (2001) identified that improved mental health was found in those MCCHD who had a stronger belief in their ability to cope and/or who used a problem-focused approach. Tak & McCubbin (2002) found that perceived social support was a predictor of parent and

family coping, such that parents who had access to use of social support as a strategy were more able to cope than those who did not.

In addition, fifteen qualitative studies identified strategies that helped PCCHD cope and build resiliency: research and becoming informed (Bratt, Jarvholm, Ekman-Joelsson, Mattson & Mellander, 2015; Bruce et al., 2014; Harvey et al., 2013; Lan et al., 2007; Sira et al., 2014); involvement in care and mastery of medical skills (Bruce et al., 2012, 2014, 2016; Lan et al., 2007; Meakins et al., 2015; Pridham et al., 2010; Sira et al., 2014); planning and taking control (Bratt et al., 2015; Bruce et al. 2014; Clark & Miles, 1999; Pridham et al., 2010; Rempel & Harrison, 2007; Sparacino et al., 1997); advocacy (Clark & Miles, 1999; Sira et al., 2014); use of the internet and social media (Bratt et al., 2015; Sira et al., 2014); supportive relationships with family/friends (Bruce et al. 2014, 2016; Lan et al.,2007; Rempel & Harrison, 2007; Sira et al., 2014); relationships with other CHD families (Sira et al., 2014; Bratt et al, 2015; Lan et al., 2007; Bruce et al. 2014, 2016); having a positive attitude, self-care, staying strong (Sira et al., 2014; Clark & Miles, 1999; Lan et al., 2007; Pridham et al., 2010; Lee & Rempel, 2011; Rempel & Harrison, 2007); celebration, acknowledgement and taking time to breathe (Sira et al., 2014; Rempel & Harrison, 2007; Redshaw, Wilson, Scarfe & Dengler, 2011; Lee & Rempel, 2011); acceptance (Meakins et al., 2015; Lan et al., 2007; Lee & Rempel, 2011); normalisation (Sira et al., 2014; Rempel & Harrison, 2007; Clark & Miles, 1999; Lee & Rempel, 2011; Rempel & Harrison, 2007; Gudmundsdottir et al., 1996; Sparacino et al., 1997) ; downplaying fears (Lee & Rempel, 2011; Rempel & Harrison, 2007); spirituality or God (Sira et al., 2014; Lan et al., 2007; Sparacino et al., 1997); formal supports such as counselling (Meakins et al., 2015; Bratt et al., 2015; Sparacino et al., 2017); telling the story (Redshaw et al. 2011; Rempel & Harrison, 2007; Gumundsdottir et al., 1996); imagining the future (Redshaw et al., 2011); self/family specific strategies (e.g. Sira et al., 2014; Lee & Rempel, 2011; Gudmundsdottir et al., 1996).

3.9.6 Unhelpful coping strategies (RQ9)

Emotion-focused coping was generally viewed as an unhelpful style of coping. Berant et al. (2001) found that PCCHS who used an emotion focused approach and/or had less belief in their ability to cope had more mental health concerns and were more likely to have attachment issues. Similarly, Berant. Mikulincer and Florian (2003) identified that the negative effects of attachment avoidance were mediated by the appraisal of motherhood in threatening terms and the reliance on emotion-focused coping.

Wray and Sensky (2004) reported that pre-operative distress was correlated with use of avoidance and waiting, and expression of emotion in mothers; and with passive patterns of reaction and palliative coping in fathers. Post-operatively, maternal distress correlated with use of a pattern of passive reaction; and for fathers with the expression of emotion. Davis et al. (1998) identified that mothers who experienced high levels of daily stress and used emotion-focused coping strategies (e.g. self-blaming, avoiding emotions) were significantly more distressed than those who did not.

Rychik et al. (2013) found that denial was most associated with traumatic stress, anxiety and depression. Doherty et al. (2009) identified that parental coping style predicted psychological distress. For mothers, 44% of the variance in psychological distress was precited by coping style (specifically behavioural disengagement), reduced understanding of the diagnosis and lack of family cohesion. For fathers, 26% of the variance in psychological distress was predicted by coping style (specifically alcohol use) and high levels of worry.

3.9.7 Interventions and coping (RQ9 and RQ10)

The efficacy of targeted interventions in increasing helpful coping strategies and resiliency in PCCHD was explored in five qualitative studies and one study that used a study-specific set of Likert scales. In a study of the use of ‘Heart Beads’ (a form of narrative therapy),

83% of mothers and 80% of fathers felt that their understanding of the cardiac condition and procedures had improved (Wilson & Chando, 2015). Similarly, Redshaw et al. (2011) found that the use of heart beads allowed parents to tell their story, acknowledge the journey and imagine the future. Further, it provided a mechanism to explain to others what had occurred.

Kocyildirim et al. (2007) reported that parents who were able to view a video of their child's heart surgery were better able to communicate their experience with family and friends, and felt more involved with their child's experience. McCrossan et al. (2008) found that providing parents with in-home medical consultations (via videoconference) increased parents' ability to seek access to medical information and to improve communication channels with the cardiac team. Finally, White et al. (2016) identified that a camp for cardiac children provided respite for parents from their identity as "cardiac" parents.

Chapter 4 - Discussion

PCCHD experience considerable psychological distress, with up to 85% of PCCHD reporting non-specific psychological distress and up to 75% of MCCHD reporting this at clinically significant levels (e.g. Bevilacqua et al., 2013; Gardner et al., 1996).

4.1 Findings Related to Research Questions

4.1.2 Manifestations of psychological distress in PCCHD (RQ1)

PCCHD manifest psychological distress through a variety of symptoms, with the symptom clusters of stress, depression, anxiety and somatisation most frequently reported at clinical levels, and long-term symptoms of anxiety, depression and somatisation also reported (e.g. Bruce et al., 2014, 2016; Clark & Miles, 1999; Lawoko & Soares, 2002, 2006; Majnemar et al., 2006; Rychik et al., 2013). Themes of sadness/loneliness; worry/concern; fear/terror; uncertainty; hopelessness; anger; insomnia; social isolation and vulnerability were also identified (e.g. Bruce et al., 2014, 2016; Carey et al., 2002; Clark & Miles, 1999; Lan et al., 2007; Pinelli et al., 1981; Pridham et al., 2010; Rempel & Harrison, 2007; Rempel et al., 2009; Sparacino et al., 1997). The symptoms of stress, depression, anxiety and somatisation are often represented as syndrome scales within broader measures of psychological distress, such as those measures reported most frequently in this review (GHQ, BSI, SCL and PSI). This may disproportionately under-represent the presence of other symptom clusters and more research is required to determine the diversity and prevalence of such symptoms for PCCHD.

4.1.2 Comparison with PHC, PCOD and normative samples (RQ2)

4.1.2.1 PHC and normative samples

In general, PCCHD reported more non-specific psychological distress, depression and anxiety than PHC and normative samples, and higher levels of depression than a psychiatric reference group (e.g. Bevilacqua et al., 2013; Brosig, Whitestone et al., 2007; Diffin et al., 2016;

Gardner et al., 1996; Lawoko & Soares, 2002, 2003a; Rahiminafar et al., 2015; 2016; Rona et al., 1998; Sklansky et al., 2003; Solberg et al., 2011b, 2012; Wray & Sensky, 2004). There were some minor exceptions to these findings, however these tended to be within studies where surgical intervention was historical (long term) and/or patients were adolescent/young adult children (e.g. Dulfer et al., 2015; Goldbeck et al., 2005).

Stress was reported to be higher than normative samples during surgery, however more research is required to determine whether PCCHD generally experience stress and somatization at levels higher or comparable to PHC (e.g. Dulfer et al., 2015; Spijkerboer et al., 2007). Qualitative responses consistently reported considerable stress in relation to psychosocial factors such as: perception of reduced child quality of life; caregiving burden; child characteristics that made the child difficult to parent; perceptions of parenting competence; challenges with discipline; reduced social support; financial pressure; family member burden; gender roles; and diagnosis/medical intervention (e.g. Brosig, Mussato et al., 2007; Brosig, Whitstone et al., 2007; Bruce et al, 2012, 2014; Cantwell-Bartl & Tibballs, 2015; Carey et al., 2002; Clark & Miles, 1999; Connor et al., 2010; Diffin et al., 2016; Franck et al., 2010; Hoehn et al., 2004; Lan et al., 2007; Lee et al., 2007; Majnemar et al., 2006; Pelchat et al., 1999; Pridham et al., 2010; Sarajuuri et al., 2010; Tak & McCubbin, 2002; Torowicz et al.,2007; Werner et al., 2014).

Together, these findings are consistent with those of existing reviews which found that psychological distress reported by PCCHD is generally at levels higher than normative samples and control groups (both for non-specific psychological distress and for specific syndrome scales; Jackson et al., 2015; Soulvie et al., 2012) but that a minority of studies show inconsistent findings (and report comparable or lower levels of psychological distress in PCCHD. Wei et al., 2015). The quantitative studies examined in this review varied greatly in relation to demographics; control factors (such as severity, surgery status, timing of diagnosis); measures

used; design; and research questions. Wei et al. (2015) suggested that such variations may have contributed to inconsistencies in findings in their review and it is likely that this was also the case for this review. Future research may benefit from use of a consistent approach and battery of measures, and future reviews may consider targeting specific measures for meta-analysis.

4.1.2.2 PCOD

PCCHD reported experiencing comparable levels of psychological distress (non-specific), stress, depression and anxiety to PCOD whose children had conditions that were more severe/chronic (bone marrow transplant, Downs Syndrome), and higher levels of anxiety and somatization than PCOD whose children had conditions that were less severe (cleft lip/palette, emergency outpatient. e.g. Arafa et al., 2008; Lawoko et al., 2002, 2003a; Pelchat et al., 1999). Similarities between PCCHD and parents of children with Downs Syndrome may be explained by the 40-60% comorbidity that people with Downs Syndrome share with CHD (Khoury & Erickson, 1992; Kim et al., 2014). Further research to explore this possibility, along with other chronic health comparisons, is recommended.

4.1.3 Differences based on timing of diagnosis (RQ3)

Consistent with the review by Soulvie et al. (2012), this systematic review found that diagnosis and birth of a child with CHD were consistently reported as periods of intense distress and anxiety for PCCHD, with PCCHD describing it as the ‘worst time of their lives’ (e.g. Brosig, Whitestone et al., 2007; Cantwell-Bart & Tibballs, 2015; Pinto et al., 2016). Prenatal diagnosis appeared to provide individuals, whose foetus had CHD (and who chose to continue with the pregnancy), an opportunity to prepare financially, logistically and emotionally for the challenges ahead (e.g. Bratt et al., 2015). However, inconsistent findings indicate that more research is required to determine whether the timing of diagnosis (prenatal versus postnatal) is related to parental psychological functioning in the short and long-term. Prenatal diagnosis tends to be

associated with more severe forms of CHD (e.g. Brosig, Whitestone et al., 2007) and it is therefore recommended that future research in this area take this into consideration in study design/analysis.

4.1.4 Differences based on the severity of the CHD (RQ4)

Severity of CHD was not found to be consistently predictive of parental psychological distress (non-specific distress and symptom scales, e.g. Berant et al., 2001, 2008; Brosig, Mussato et al., Goldbeck and Melches, 2006; 2007; Solberg et al., 2011a, 2011b, 2012; Yildiz et al., 2009). At face value, this finding contradicts that of the Jackson et al. (2015) review which found that CHD complexity was consistently associated with increased psychological distress for PCCHD. However, a more detailed examination of Jackson et al.'s review identifies a number of risk factors for parents of children with complex CHD, that are consistent with those found in this review (and those identified by the 2012 Soulvie et al. review). Specifically, risk factors such as: prolonged hospitalization; repeated surgeries; perceptions of reduced child quality of life; increased caregiving burden; financial difficulties; parenting concerns; and social isolation (e.g. Berant et al., 2001, 2008; Goldbeck and Melches, 2006; Lawoko & Soares, 2004, 2006; Sarajuuri et al., 2012; Werner et al., 2014). It seems likely that the relationship between CHD severity and parental distress may not be causal in nature but a more complicated one in which parents whose children have severe CHD are more likely to experience risk factors for psychological distress and therefore more likely to experience increased psychological distress. These findings are also consistent with those of Tedstone and Tarrier (2003), who found that psychological symptoms (in post-treatment adult medical patients) are not usually predicted by illness severity but by other factors such as negative appraisal and poor social support.

Soulvie et al. (2012) identified few longitudinal studies in their systematic review. However, approximately one third of studies included in this review utilized a repeated measures

design, and 11.5% reported on data collected over twelve months or more (e.g. Berant et al., 2003, 2008; Diffin et al., 2016; Solberg et al., 2011b, 2012). In general, these studies tended to highlight a different trajectory of symptoms when CHD was severe, and differences in symptom manifestation based on the timing of the measure. Taken together, these findings suggest that cross-sectional research may need to be applied with caution as this may not adequately capture the level of distress experienced by PCCHD at different points in time, nor capture the development of chronic mental ill-health in PCCHD. Many of the studies in this paper employed a prospective cohort approach and it will be interesting to see research findings as these cohorts are tracked into the future.

4.1.5 Differences based upon surgical intervention (RQ5)

Surgery (decision making, preparation, process and recovery) was consistently reported as a period of high stress for PCCHD (e.g. Cantwell-Bartl & Tibballs, 2015; Clark & Miles, 1999; Connor et al., 2010; Diffin et al., 2016; Franck et al., 2010; Lan et al., 2007). Whilst consistently high levels of psychological distress (up to 82%), anxiety and somatization were found in parents whose children had experienced cardiac interventions (particularly when the intervention was more invasive, e.g. open-heart surgery), this was shown to decrease over time (anxiety and psychological distress only) following surgery (e.g. Bevilaqua etal., 2013; Landolt et al., 2011; Utens et al., 2002; Vrijmoet-Wiersma et al., 2009; Wray & Sensky, 2004). Depression tended to be higher when children required cardiac surgery rather than catherization (e.g. Diffin et al., 2016; Solberg et al., 2011b, 2012; Utens et al., 2002).

These findings are consistent with the review by Soulvie et al. (2012) who found that diagnosis and treatment are consistent stressors for PCCHD. Taken together, findings suggest that CHD severity (in a medical sense) was not as salient for the experience of distress, as was the initial diagnosis and period of surgical intervention (e.g. Cantwell-Bartl & Tibballs, 2015;

Clark & Miles, 1999; Connor et al., 2010; Diffin et al., 2016; Franck et al., 2010; Lan et al., 2007). Further, the requirement for open-heart surgery (as opposed to catherization, and regardless of surgical complexity) appeared to be, in and of itself, distressing, with successful completion associated with at least a temporary period of reduced distress (e.g. Bevilaqua et al., 2013; Diffin et al., 2016; Landolt et al., 2011; Utens et al., 2002; Vrijmoet-Wiersma et al., 2009; Wray & Sensky, 2004).

One explanation for this, is that for non-medical PCCHD, open-heart surgery is likely to be viewed as a pervasive experience of uncertainty with the constant threat of the child's death (Rempel et al., 2012). More specifically, qualitative and quantitative studies that reflect upon distress associated with the sights and sounds of ICU (as well as the behaviour of the infant or child in ICU post-surgery), and studies that show a relationship between parental distress and perceptions of their child's quality of life, indicate that parental distress may be influenced by their appraisal that open-heart surgery is serious, that their child may die, and that their child will suffer to some degree (regardless of the complexity of either the condition or the surgery itself; e.g. Carey et al., 2002; Clark & Miles, 1999; Cantwell-Bart &Tibballs, 2015; Connor et al., 2010; Diffin et al., 2016; Franck et al., 2010; Harvey et al., 2013; Kocyilidrim et al.; 2007; Lee & Rempel, 2011; Meakins et al., 2015; Rempel & Harrison, 2007; Sparacino et al., 1997).

This experience may also be exacerbated by lack of familiarity with medical perspectives of surgery (where risk is focused more on the complexity of procedure). In a qualitative analysis of parental reactions to being shown footage of their child's cardiac surgery, Kocyilidirin et al. (2007) found that this experience was important in helping parents to understand what their child had experienced and to fill the gaps in time when they were unable to be with their child (surgery often being the only time where they were separated). Interestingly, the only parent not reporting this same benefit, was a health professional with experience in surgical settings. This explanation

is, to some degree, speculative in nature and further research is recommended to explore this. Qualitative approaches that focus on meaning finding may be particularly suitable.

4.1.6 Differences based on parent gender (RQ6)

Consistent with existing reviews (Jackson et al.,2015; Soulvie et al., 2012; Wei et al., 2015), studies in this review more often concerned the experiences of MCCHD (30.5%) than FCCHD (3.7%), although almost two thirds of studies (65.9%) included both. MCCHD consistently reported greater levels of psychological distress (non-specific), depression, anxiety and somatization than FCCHD (e.g. Bevilacqua et al., 2013; Doherty et al., 2009; Hoehn et al., 2004; Lawoko & Soares, 2002, 2003a, 2006; Pinto et al., 2016; Utens et al., 2002; Wray & Sensky, 2004; Yildiz et al., 2009). In general (but with some exceptions), similar levels of stress were experienced by MCCHD and FCCHD, however the nature of this stress differed between them (e.g. Franck et al., 2010; Sarajuuri et al., 2012; Werner et al., 2014). These findings indicate that future research into psychological distress would benefit from controlling for gender.

Qualitative reports consistently identified that psychological distress was experienced by both MCCHD and FCCHD (albeit sometimes in unique ways) and this may suggest that the contextualisation of distress is an important feature for being able to accurately identify and then measure it (e.g. Franck et al., 2010; Hoehn et al., 2004). Of particular interest, within this context, was the experience of ‘role loss’ described by both mothers and fathers (e.g. Bruce et al.,2016; Diffin et al 2016;). For MCCHD, the inability to take part in basic caring tasks, such as feeding, washing, cuddling and soothing their child (tasks traditionally associated with the role of mother) was a source of distress; and for FCCHD, a dichotomy existed between a task-oriented traditional role of working to provide for the family (and the difficulties in undertaking this whilst the child was in hospital) and changes in gender roles (in some cultures) where fathers

sought out a more direct caregiving role but felt denied in this by both gender expectations and by hospital restrictions (e.g. Bruce et al., 2016; Clark & Miles, 1999; Diffin et al 2016;).

Exploration of this phenomenon requires further research into changes in gender and parenting identity; differences in this experience within cross-cultural contexts; and the interface of this within the context of CHD and medical models of care. Consideration should be given as to whether existing measures of psychological distress adequately address the idioms of distress exhibited by MCCHD and FCCHD in relation to specific stressors and cultural constructs of parenting identity, and whether these factors are predictive of distress in a way that is modifiable. In addition, the suggestion that FCCHD may experience intense emotion but feel pressure to appear strong (Clark & Miles, 1999) raises the possibility that FCCHD may under-report psychological distress.

4.1.7 The experience of trauma-related symptoms (RQ7)

A relatively small number of quantitative studies investigated the experience of trauma in relation to PCCHD (n=4) and consistently reported that PCCHD experience trauma symptoms at clinically significant levels, with up to 26% found to meet full criteria for PTSD; up to 34% to meeting criteria for Acute Stress Disorder; and up to 83% having at least one clinically significant symptom (with most having multiple symptoms. Franich-Ray., 2012; Hefricht et al., 2009; Landolt., 2011; Rychik et al., 2013). Helfrict et al. (2009) also found that the prevalence of Acute Stress Disorder was 6.25 times higher in PCCHD than in adult cardiac patients. Whilst the study of trauma in relation PCCHD is an under-developed area of research, the findings nevertheless concur with existing research into the trauma experiences of parents in relation to oncology settings (Kohlsdorf & Costa Jnr, 2012), where such parents were found to be at increased risk of PTS (up to 99% prevalence for symptoms and 30% for PTSD. e.g. Karadeniz et al., 2017; Kazark et al., 2004 & 2005; Ljungman et al., 2015; Stuber et al., 1996). These findings

are also consistent with other studies that have found PTSD, PTSS and Acute Stress Disorder in parents exposed to child-related trauma such as non-fatal accidents; disability; chronic illness; and/or medical interventions (e.g. de Vries et al., 1999; Balluffi et al., 2004; Kazak et al., 2004; LeGouez et al., 2016; Robert et al., 2014).

Additional support for the finding that PCCHD experience trauma-related symptoms, came from 11 studies (13.41%), which identified the following themes: flashbacks; hypervigilance; dissociation; avoidance; constant fear that the child would die; vivid recounting of the sights/sounds of NICU and the behaviour/appearance of the child post-surgery (e.g. Carey et al., 2002; Clark & Miles, 1999; Diffin ey al., Franich-Ray et al., 2012; Lee & Rempel, 2011; Meakins et al., 2015; Rempel & Harrison, 2007; Rychik et al., 2013; Sparacino et al., 1997). Experiences reported in these studies were also recounted in ways consistent with PTSD Criterion A2 of the Diagnostic and Statistical Manual of Mental Disorders, fourth edition (DSM-IV): fear (‘terrified’), helplessness (‘loss of control’, ‘uncertainty’), and horror (‘devastation’, ‘anguish’, ‘appearance of child’).

The inclusion of qualitative studies in this review, was in response to the recommendations of Wei et al. (2015) and Soulvie et al. (2012), who argued that narrow response options in quantitative studies do not provide adequate scope for meaning-finding, and that future studies of the experience of CHD families would benefit from the integration of qualitative and quantitative material. In this case, the exploration of trauma benefitted from this approach. Further research is required to establish a more extensive evidence-base for the existence of trauma-related symptoms in PCCHD and, in keeping with Soulvie et al’s recommendation, an integrated mixed methods approach to this is recommended.

4.1.7.1 Hypervigilance

This review found that symptoms of hypervigilance were consistently reported by PCCHD (e.g. Meakins et al., 2005; Sparacino et al., 1997; White et al., 2016). Hypervigilance was used to deal with uncertainty and to attempt to gain control of a situation that was not able to be controlled (e.g. Lee & Rempel., 2011; Meakins et al., 2015; Pridham et al., 2010). Hypervigilance also appeared to have originated in an appropriate need for medical vigilance (e.g. care of a medically fragile infant) but was a practice that did not reduce as the threats to the child became less salient (e.g. increased stability of the child's health or the ability of the child to participate in their own health care. Meakins, et al., 2015; Sparacino et al., 1997; White et al., 2016). This finding is consistent with Kimble et al. (2013) who found exposure to a traumatic situation (war-zone deployment) for military personnel (who were trained to be vigilant), predicted the development of PTSD but also, independently, hypervigilance. Kimble et al. (2013) cautioned that hypervigilance should therefore only be characterised as indicative of PTSD when taken in the context of the individual experience and the presence of other symptoms. Within the context of PCCHD, the child's diagnosis and experience of surgery/medical interventions, along with evidence that indicates the presence of PTSS, suggest that hypervigilance may be indicative of PTSD. Further research is required to explore this hypothesis.

4.1.7.2 Symptoms of avoidance

Helfricht et al. (2009) suggested that measures of trauma that define avoidance in active terms (i.e. physical avoidance) may not be best suited to studies of PCCHD. More specifically, PCCHD may not be able to engage in physical avoidance as a coping strategy (e.g. through the need to attend medical appointments etc) but may use more passive forms of avoidance (e.g. numbing/ emotional distancing) instead. Findings of avoidant-attachment in MCCHD post-diagnosis (Berant et al., 2001), and accounts of parental fear of attachment based upon the

possibility of their child's mortality (Clark & Miles, 1999), are supportive of this hypothesis. These findings are consistent with Norberg and Boman (2009) who found that parents within oncology settings often express initial PTSS as avoidance. Further research to explore this is recommended.

4.1.7.3 Comorbidity of symptoms

The Transdiagnostic Model suggests that common risk factors may lead to multiple disorders (multifinality), and that these may co-exist or develop in differing ways depending on the individual (divergent trajectories. e.g. Nolen-Hoeksema & Watkins, 2011). Research indicates that trauma symptomology shows significant overlap with those of other conditions, such as anxiety and depression (e.g. Flory & Yehuda., 2015; Spinhoven, Penninx, van Hemert, de Rooij & Elzinga, 2014). For example, using the four-factor model of PTSD (dysphoria, re-experiencing, avoidance and hyperarousal. Simms et al., 2002), research has shown that depression is highly correlated with the dysphoria found in PTSD (Simms et al., 2002) and PTSD and Major Depressive Disorder (MDD) share a 50% comorbidity (Flory & Yehuda, 2015). Despite this overlap, PTSD and depression have been shown to be best understood as separate constructs that share symptoms of distress and co-occur frequently (Post, Zoellner, Youngstrom & Feenva, 2011). The comorbidity of PTSS and symptoms of psychological distress (generic and symptom specific) has been more specifically investigated in parents whose children were admitted to PICU for life-threatening injuries/illnesses. Findings report that up to 54% met clinically significant criteria for Acute Stress Disorder; 27% for depression and anxiety; and 31% for stress. Whilst symptoms were shown to significantly correlate, they did not vary as a function of child diagnosis (which included CHD; Muscara et al., 2015).

Similarly, a number of other studies included within this review showed some evidence of symptom comorbidity (e.g. Lawoko & Soares,2002, 2003a, 2004, 2006; Solberg et al., 2011a,

2011b, 2012). Whilst none examined PTS within this context, evidence supporting the existence of PTS in PCCHD, along with evidence of comorbidity of other symptoms, suggests that PCCHD are likely to experience trauma and psychological distress in much the same way as parents of children with other significant health concerns. Research that explores comorbidity of PTSS and other symptoms of psychological distress within the PCCHD context is therefore recommended.

4.1.7.4 Specificity of measures

Studies within this review tended to include broad measures of psychological distress to identify symptoms of psychological distress, relying on symptom subscales of each measure to determine syndromes (e.g. Bevilaqua et al., 2013; Dulfer et al., 2015; Spijkerboer et al., 2007). For example, the GHQ-12 & 28 calculate subscales of somatisation; anxiety/insomnia; social dysfunction; and severe depression, but do not specify a trauma subscale. Research has demonstrated, however, that broader measures of psychological functioning, such as the GHQ, may actually be effective screening tools for PTS (e.g. Ouimette, Wade, Prins & Schohn, 2007). Therefore, the studies within this review that used such measures for PCCHD, may also contain valuable trauma-related data that has not yet been explored or reported on within this context.

In contrast, other measures may lack the scope to adequately screen for PTS by solely capturing comorbid (but not unique) PTSS. In these cases, extrapolating findings to report on the prevalence of mental-ill-health in general for PCCHD, may be susceptible to under-reporting. More specifically, it may not capture those PCCHD whose symptoms are more unique of PTS than symptom syndromes captured by these measures (e.g. Steel, Dunlavy, Stillman & Pape, 2011). If this is the case, it may explain some of the inconsistencies in quantitative findings found in this review. Future research involving measures carefully selected to discriminate PTS in PCCHD is recommended (Steel et al. 2011).

4.1.7.5 Symptom trajectories

This review found evidence to suggest that trajectories of symptoms may vary as a function of time and symptom type, with some symptoms evident in the short-term and others more apparent over time (e.g. e.g. Berant et al., 2003, 2008; Diffin et al., 2016; Solberg et al., 2011b, 2012). This finding is consistent with research into PCOD, particularly those in NICU situations, where there are variations between and within symptoms of PTS and other forms of psychological distress over time (e.g. Greene et al., 2015; Baluffi et al., 2004; Ljungman et al., 2015). Norberg and Boman's (2009) research into PTSS in oncology settings, identified a period of initial high-level-distress that tended to ameliorate over time, but with an exacerbation of initial symptoms of PTS (such as avoidance) into chronic PTSD over time. Berant et al., (2001) found that maternal attachment avoidance following infant diagnosis was related to poor mental health at diagnosis, deterioration of mental-health over a twelve-month period and poorer mental health seven years later. Whilst not reported within the context of trauma-related symptoms, this trajectory shows some consistency with the findings of Norberg and Boman.

Further, the findings of Lawoko & Soares (2002, 2003a, 2004, 2006) and Solberg et al. (2011a, 2011b, 2012) suggest that elevated levels of depression and anxiety may continue to be apparent for up to three years. Taken within the context of comorbidity of PTS with anxiety and depression, these findings may indicate that long-term PTS is evident in PCCHD. The importance of future research to address these questions is further emphasised by Berant et al.'s (2001, 2003, 2008) finding that maternal avoidant attachment, along with related long-term maternal mental ill-health, is associated with emotional problems and poor self-image in CHD children at seven years post-diagnosis.

4.1.8 The experience of Post-Traumatic Growth (RQ8)

This review did not find any studies that specifically examined, nor defined findings in the context of, PTG. However, a number of studies provided accounts suggestive of the transformative nature of the CHD experience for PCCHD. More specifically, themes were identified that related to benefits and blessings; gratitude, appreciation and re-prioritisation; spiritual connection and meaning finding; mastery and pride; and 'paying it forward' (e.g. Brosig, Mussator et al., 2007; Bruce & Sundin, 2012; Bruce et al., 2014; Carey et al., 2002; Clark & Miles, 1999; Harvey et al., 2013; Jordan et al., 2014; Kocyildirim et al., 2007; Lan et al., 2007; Lee & Rempel, 2011; Meakins et al., 2015; Rempel & Harrison, 2007; Rempel et al., 2009; Sira et al., 2014; Sparacino et al., 1997). These themes are consistent with definitions of PTG found in previous research, specifically: thriving; spiritual connection or existential comprehension; sense of stronger self; re-prioritisation of what is considered important in life; and/or more meaningful relationships with others (Calhoun & Tedeschi, 1999, 2001; Carver, 1998; Tedeschi & Calhoun, 1996, 2004). These findings are also consistent with research that found evidence of PTG in parents of children with life-limiting/threatening conditions (e.g. Turner-Sack et al., 2016); and with research that found PTG in 54.3% of parents of children undergoing corrective surgery for congenital defects (Li et al., 2012).

In addition, consistent with research that identifies PTG and PTS as separate but commonly co-occurring phenomena, and research that identifies some overlap between PTG and coping/resiliency (Armstrong, Shakespeare-Finch & Shochet, 2014; Shakespeare-Finch & Lurie-Beck, 2014; Tedeschi & Calhoun, 2004), a number of studies within this review shared reports from PCCHD that expressed elements of both PTS and PTG (Carey et al., 2002; Clark & Miles, 1999; Harvey et al., 2013; Lee & Rempel, 2011; Meakins et al., 2015; Rempel & Harris, 2007; Sparacino et al., 1997) and PTG and coping (Clark & Miles, 1999; Diffin et al., 2016; Harvey et

al., 2013; Meakins et al., 2015; Prodham et al., 2010; Rempel & Harrison, 2007; Rychik et al., 2013; Sparacino et al., 1997; White et al., 2016), with some sharing features of all three (Clark & Miles., 1999; Harvey et al., 2013; Meakins et al., 2015; Rempel & Harrison; 2007; Spacino et al., 1997) . Taken together this suggests that PCCHD may experience PTG in ways that not only allow them to transcend their experience in some way but also to actively 'give back' to the community. This finding is a unique contribution of this review and future research targeting PTG in PCCHD is recommended. Mixed design studies that effectively integrate qualitative and quantitative findings may be particularly applicable, as might those designed to investigate the co-occurrence of PTG wand PTS, and the relationship of these to resiliency and coping strategies in PCCHD.

4.1.9 Interventions and experiences of coping (RQ9 and RQ10)

4.1.9.1 Coping (RQ9)

Qualitative studies identified a number of strategies that helped PCCHD to cope and build resiliency, including: research and becoming informed; involvement in care and mastery of medical skills; planning and taking control; advocacy; use of the internet and social media; supportive relationships with family/friends; relationships with other CHD families; having a positive attitude, self-care, staying strong; celebration, acknowledgement and taking time to breathe; acceptance; normalisation; downplaying fears; spirituality or God; formal supports such as counselling; telling the story; imagining the future; and self/family specific strategies (Bratt et al., 2015; Bruce & Sundin, 2012; Bruce et al., 2014, 2016; Clark & Miles, 1999; Gudmundsdottir et al., 1999; Harvey et al., 2013; Lee & Rempel, 2011; Lan et al., 2007; Meakins et al., 2015; Pridham et al., 2010; Redshaw et al. 2011; Rempel & Harrison, 2007; Sira et al., 2014; Sparacino et al., 1997). The coping strategies identified were generally reported in highly personal ways, reinforcing the individuality of coping mechanisms and the importance that individual context

has in this process. Considered together with the wide array of risk factors identified for psychological distress, these findings highlight the importance of individual family-centered screening and psychosocial interventions for families of children with CHD.

Consistent with the findings of existing reviews (Jackson et al., 2015; Soulvie et al., 2012; Wei et al., 2015), quantitative studies of coping varied greatly in the research questions investigated and this made it difficult to draw general conclusions from the findings. Nevertheless, similar to existing research into chronic illness, PCCHD who used emotion-focused coping styles, avoidance/denial, wishful thinking and self-blame, were more likely to have reduced psychological functioning; and those that used medical information seeking, medical communication and spirituality, tended to report greater ability to cope (e.g. Berant et al., 2001; Davis et al., 1998; Rychik et al., 2013; Sira et al., 2014; Wray & Sensky, 2004).

4.1.9.2 Interventions (RQ10)

Re et al. (2011) identified that the most effective way to improve infant mental health after cardiac surgery was to treat MCCHD psychological distress. Given the breadth of evidence within systematic reviews that indicates PCCHD experience considerable amounts of non-specific and symptom-specific psychological distress (Jackson et al., 2015; Soulvie et al., 2012; Wei et al., 2015; and the findings of this review) and evidence that suggests that parental well-being impacts on the physical and mental health of their children (e.g. Berant et al., 2008; Re et al., 2011)**,** the number of studies evaluating interventions for PCCHD appears to be disproportionately low (n=6;). It is unclear, however, whether this is reflective of few evaluations, or of low intervention rates. Nevertheless, of the six interventions included in this review, all showed some success in reducing distress in PCCHD and promoting resiliency/positive coping strategies (e.g. McCusker et al., 2010; McCrossan et al., 2008).

Whilst interventions appeared to be mostly localised (health service specific), all focused on ways to: address anxiety/vigilance (e.g. respite; information provision; skills training; timely access to medical consultation); include PCCHD in the experiences of their children (e.g. videos of surgery; heart beads; presence at anesthetic); and/or integrate these experiences into their own narrative of the CHD journey (e.g. videos of surgery, heart beads. Kocyildirim et al., 2007; McCrossan et al., 2008; McCusker et al., 2012; Odegard et al., 2002; Redshaw et al., 2011; White et al., 2016; Wilson & Chando et al., 2015). Interestingly, this focus upon the reduction of anxiety and hypervigilance, and the promotion of a sense of control (through the building of emotional and CHD-specific competencies), appears to be targeting symptoms consistent with the experience of PTS (as well as comorbid anxiety and depression). The development, piloting and evaluation of further intervention programs for PCCHD (along with the publication of results) is strongly recommended, as is the consideration of parent well-being when paediatric hospital units are being designed and/or refurbished. Such interventions need not be costly in nature but can involve the rethinking of existing and future resources to remove unnecessary obstacles to participation in care; reduce environmental restraints; improve early psychosocial screening and family-centred care; and explore increased partnerships with non-for-profit organisations to provide psychosocial support to PCCHD on-site. The use of peer supports and online support programs, may also have potential as cost-effective strategies for the reduction of social isolation.

4.2 Limitations of the Current Review

In an attempt to capture as many published studies as possible relating to the topic of this review, restrictions were not placed on studies based upon the measures used, design nor year of publication. Soulvie et al. (2012) and Wei et al. (2015) have indicated that the integration of qualitative and quantitative findings provides a richer understanding of parental experiences, and

this approach was applied within this review. However the resulting integration of evidence into a cohesive argument was subject to some challenges. For example, the volume of included studies, and the pursuit of parsimony, meant that analyses regarding country of origin, sample size and type of measures used, was not possible. Further, medical interventions and treatments for CHD are continually evolving, and childhood survival rates have increased dramatically over time. By placing no restrictions on the year of publication for studies included in this review, it is possible that this may have confounded findings to some degree. Nevertheless, the finding that CHD complexity may not directly increase psychological distress, along with the finding that it is psychosocial stressors that may more readily predict such distress, indicates that the year of publication (as a reflection of distress related to less sophisticated medical interventions and increased mortality rates) may not be as salient.

4.3 Summary

This systematic review was designed to explore the experience of psychological distress, PTS, PTG and coping in PCCHD. Consistent with the findings of existing reviews (Jackson et al., 2015; Soulvie et al., 2012; Wei et al., 2015), this review found that PCCHD experience considerable distress that manifests in a variety of symptoms (including depression, anxiety, stress and somatization). This distress is generally higher than normative populations and is consistent with that experienced by PCOD (chronic and severe). The experience of distress tends to be exacerbated (or precipitated) by a number of psychosocial stressors and these are more commonly found in families where the CHD is more severe in nature. Birth, diagnosis, and invasive surgical procedures (open heart surgery) are a significant source of distress for PCCHD, regardless of CHD severity or the complexity of the surgical procedure. Consistent with findings into trauma reactions in PCOD, these events may service as a trigger for parental concerns about child mortality and suffering, and the experiences of role loss, uncertainty and lack of control. A

number of PCCHD met the diagnostic threshold for Acute Stress Disorder and Post-Traumatic Stress Disorder, with many more experiencing multiple clinically significant symptoms. Ongoing symptoms of trauma (especially hypervigilance), depression and anxiety were also frequently reported. These trauma-related findings are a new contribution to reviews of the psychological experiences of PCCHD. In addition, PCCHD employ a variety of coping strategies to deal with their experiences, whilst also appearing to experience the transformational nature of trauma in a way that is consistent with the experience of PTG and findings relating to PTG in PCOD. This finding, to the best knowledge of the author, is a unique contribution to the field and has not been explored in either single studies or reviews of PCCHD.

Together these findings highlight a number of key areas for future research within the PCCHD population, including longitudinal studies to determine the range, prevalence and trajectory of psychological distress, PTSS and PTG; the development and/or identification of effective and consistent screening instruments to discriminate PTS from other forms of psychological distress (and to identify the existence of, and potential for, PTG); the development, piloting and implementation of interventions to reduce parental anxiety and to increase parent resilience, healthy coping, sense of control and inclusion; qualitative longitudinal studies to explore the meaning behind parental distress (especially in relation to surgery) and the experience of PTG; use of consistent methodology/tools to improve inter-study comparison; the exploration of factors related to gender identity and cultural idioms of distress; and the piloting and evaluation of interventions designed to reduce symptoms of psychopathology, build resiliency and coping and promote PTG. Finally, this paper highlighted the way in which research into the psychological experiences of PCCHD can be used to better inform public policy in healthcare settings, not only to reduce psychosocial morbidity for PCCHD but to also improve the physical and mental health outcomes of children with CHD.

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Appendix A

Tables A1 and A2

Table A1

Common Terms and Definitions

Terms	Definitions
Acyanotic	Normal blood oxygen saturation
Anxiety symptoms	Symptoms characteristic of a psychiatric diagnosis of anxiety. May include full or partial symptoms
Bi-Ventricle Physiology	Heart conditions where both ventricles are present/working
Cardiac catheterisation	Cardiac investigations or interventions performed using a thin tube inserted into a large blood vessel and threaded into the heart
Congenital Heart Disease (CHD)	Structural or functional malformation of the heart, valves or central blood vessels present at birth (for a detailed breakdown, see Table 1.1)
Cyanotic	Reduced blood oxygen saturation, usually clinically evident at <85%
Depressive symptoms (or symptoms of depression)	Symptoms characteristic of a psychiatric diagnosis of depression. May include full or partial symptoms
Open-heart surgery	Heart surgery requiring the opening of the chest to expose the heart and the placing of the blood on a heart-lung bypass machine
Palliative surgery	Surgery performed to improve functioning/alleviate pain but without complete repair
Post-Traumatic Stress	Psychological dysfunction experienced in relation to a traumatic experience that is consistent with symptoms found in PTSD or Acute Stress Disorder
Post-Traumatic Growth	Positive psychological response to the experience of trauma in which an individual not only recovers a previous level of functioning but is also able to transcend this in some way
Psychological Distress	Non-specific (or general) symptoms of psychiatric dysfunction indicating reduced mental health and wellbeing (may encompass symptoms of depression, anxiety, somatisation, stress, anger, insomnia, social isolation etc)
Single Ventricle Physiology	Heart conditions involving only a single working ventricle
Somatisation	Physical manifestation of psychological distress, where the bodily symptoms have no medical basis
Stress	Symptoms consistent with the experience of being psychological overwhelmed to some degree. Distinguishable from depression, anxiety and somatisation as a discreet subset of psychological distress
Trauma	Distressing of life-threatening event

Table A2

*Abbreviations**

Abbreviation	Term
AoS	Aortic Stenosis
ASD	Atrial Septal Defect, Acute Stress Disorder
AVS	Aortic Valve Stenosis
AVSD	Atrioventricular Septal Defect
BVP	Bi-Ventricle physiology
CAVC	Complete Atrioventricular Canal Defect
CoA	Coarction of the Aorta
COARC	Coarction of the Aorta
CHD	Congenital Heart Disease
DORV	Double Outlet Right Ventricle
DS	Down Syndrome
FCCHD	Fathers or children with congenital heart disease
HLHS	Hypoplastic Left Heart Syndrome
IAA	Interrupted Aortic Arch
ICU	Intensive Care Unit
JBI	Joanna Briggs Institute
MCCHD	Mothers of children with congenital heart disease
NHMRC	National Health and Medical Research Council
NICU	Neonatal Intensive Care Unit
NOS	Newcastle-Ottawa Scale
OCEBM	Oxford Centre for Evidence Based Medicine
PCCHD	Parents of children with congenital heart disease
PCOD	Parents of children with other disorders
PDA	Patent Ductus Arteriosus
PHC	Parents of healthy children
PICU	Pediatric Intensive Care Unit
PS	Pulmonary Stenosis
PTG	Post-Traumatic Growth
PTSD	Post-Traumatic Stress Disorder
PTS	Post-Traumatic Stress
PTSS	Post-Traumatic Stress Symptoms
PVS	Pulmonary Valve Stenosis
SVP	Single ventricle physiology
TA	Truncus Arteriosus
TAPVC	Total Anomalous Pulmonary Venous Connection
TAPVR	Total Anomalous Pulmonary Venous Return
TGA	Transposition of the Greater Arteries
ToF	Tetralogy of Fallot
UVH	Uni-Ventricular Heart
VSD	Ventricular Septal Defect

*Abbreviations for measures are in Table M1 (Appendix M).

Appendix B

Newcastle-Ottawa Quality Assessment Scale for Cohort Studies^

NEWCASTLE - OTTAWA QUALITY ASSESSMENT SCALE: COHORT STUDIES

Note: A study can be awarded a maximum of one star for each numbered item within the Selection and Outcome categories. A maximum of two stars can be given for Comparability

Selection

1) Representativeness of the exposed cohort
a) truly representative of the average _____ (describe) in the community ✸
b) somewhat representative of the average _____ in the community ✸
c) selected group of users eg nurses, volunteers
d) no description of the derivation of the cohort

2) Selection of the non exposed cohort
a) drawn from the same community as the exposed cohort ✸
b) drawn from a different source
c) no description of the derivation of the non exposed cohort

3) Ascertainment of exposure
a) secure record (eg surgical records) ✸
b) structured interview ✸
c) written self report
d) no description

4) Demonstration that outcome of interest was not present at start of study
a) yes ✸
b) no

Comparability

1) Comparability of cohorts on the basis of the design or analysis
a) study controls for the most important factor✸
b) study controls for any additional factor ✸

Outcome

1) Assessment of outcome
a) independent blind assessment ✸
b) record linkage ✸
c) self report
d) no description

2) Was follow-up long enough for outcomes to occur
a) yes (select an adequate follow up period for outcome of interest) ✸
b) no

3) Adequacy of follow up of cohorts
a) complete follow up - all subjects accounted for ✸
b) subjects lost to follow up unlikely to introduce bias or description provided of those lost) ✸
c) follow up rate inadequate and no description of those lost
d) no statement

^ Reproduced from: Wells et al. (2014).

Appendix C

The Modified Newcastle Ottawa Scale for Cross-Sectional Studies^

Selection: (Maximum 5 stars)

1) Representativeness of the sample:

a) Truly representative of the average in the target population. * (all subjects or random sampling)

b) Somewhat representative of the average in the target population. * (non-random sampling)

c) No description of the sampling strategy.

2) Selected group of users

a) Due to relevant selection of individuals to exclude factors that will bias results (such as certain diseases or drugs that have an negative/positive effect on bones) *

b) No relevant/systematic selection

3) Sample size:

a) Justified and satisfactory (power calculation included). *

b) Not justified.

4) Diagnose:

a) Characterization of the diagnosis of diabetes subtype **

b) Diabetes subtype is provided *

c) No information regarding diabetes subtype

Comparability: (Maximum 2 stars)

1) The subjects in different outcome groups are comparable, based on the study design or analysis. Confounding factors are controlled.

a) The study controls for the most important factor (select one). *

b) The study control for any additional factor. *

Outcome: (Maximum 3 stars)

1) Ascertainment of the method:

a) Validated measurement method (interassay CV included). **

b) Non-validated measurement method, but the method is available or described. *

c) No description of the measurement tool.

2) Statistical test:

a) The statistical test used to analyze the data is clearly described and appropriate, and the measurement of the association is presented (including SD/SE and the probability level (p value)). *

b) The statistical test is not appropriate, not described or incomplete.

Reproduced from: Modesti et al. (2016)

Appendix D

Oxford Centre for Evidence-Based Medicine, 2011 Levels of Evidence^

Oxford Centre for Evidence-Based Medicine 2011 Levels of Evidence

Question	Step 1 (Level 1*)	Step 2 (Level 2*)	Step 3 (Level 3*)	Step 4 (Level 4*)	Step 5 (Level 5)
How common is the problem?	Local and current random sample surveys (or censuses)	Systematic review of surveys that allow matching to local circumstances**	Local non-random sample**	Case-series**	n/a
Is this diagnostic or monitoring test accurate? (Diagnosis)	Systematic review of cross sectional studies with consistently applied reference standard and blinding	Individual cross sectional studies with consistently applied reference standard and blinding	Non-consecutive studies, or studies without consistently applied reference standards**	Case-control studies, or "poor or non-independent reference standard**	Mechanism-based reasoning
What will happen if we do not add a therapy? (Prognosis)	Systematic review of inception cohort studies	Inception cohort studies	Cohort study or control arm of randomized trial*	Case-series or case-control studies, or poor quality prognostic cohort study**	n/a
Does this intervention help? (Treatment Benefits)	Systematic review of randomized trials or *n*-of-1 trials	Randomized trial or observational study with dramatic effect	Non-randomized controlled cohort/follow-up study**	Case-series, case-control studies, or historically controlled studies**	Mechanism-based reasoning
What are the COMMON harms? (Treatment Harms)	Systematic review of randomized trials, systematic review of nested case-control studies, *n*-of-1 trial with the patient you are raising the question about, or observational study with dramatic effect	Individual randomized trial or (exceptionally) observational study with dramatic effect	Non-randomized controlled cohort/follow-up study (post-marketing surveillance) provided there are sufficient numbers to rule out a common harm. (For long-term harms the duration of follow-up must be sufficient.)**	Case-series, case-control, or historically controlled studies**	Mechanism-based reasoning
What are the RARE harms? (Treatment Harms)	Systematic review of randomized trials or *n*-of-1 trial	Randomized trial or (exceptionally) observational study with dramatic effect			
Is this (early detection) test worthwhile? (Screening)	Systematic review of randomized trials	Randomized trial	Non -randomized controlled cohort/follow-up study**	Case-series, case-control, or historically controlled studies**	Mechanism-based reasoning

*** Level may be graded down on the basis of study quality, imprecision, indirectness (study PICO does not match questions PICO), because of inconsistency between studies, or because the absolute effect size is very small; Level may be graded up if there is a large or very large effect size.**

**** As always, a systematic review is generally better than an individual study.**

^Reproduced from: OCEBM Working Group (2011)

Appendix E

*JBI Critical Appraisal Checklist for Qualitative Research^*

JBI Critical Appraisal Checklist for Qualitative Research

Reviewer_____Date_____

Author _____Year_____Record Number_____.

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

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

^Reproduced from: JBI (2014a)

Appendix F

JBI Levels of Evidence for Meaningfulness^

LEVELS OF EVIDENCE FOR MEANINGFULNESS

1. Qualitative or mixed-methods systematic review
2. Qualitative or mixed-methods synthesis
3. Single qualitative study
4. Systematic review of expert opinion
5. Expert opinion

^Reproduced from: JBI (2014b)

Appendix G

*JBI Critical Appraisal Checklist for Randomised Control Trials^*

JBI Critical Appraisal Checklist for Randomized Controlled Trials

Reviewer_____Date_____

Author _____Year_____Record Number_____

		Yes	No	Unclear	NA
1.	Was true randomization used for assignment of participants to treatment groups?	☐	☐	☐	☐
2.	Was allocation to treatment groups concealed?	☐	☐	☐	☐
3.	Were treatment groups similar at the baseline?	☐	☐	☐	☐
4.	Were participants blind to treatment assignment?	☐	☐	☐	☐
5.	Were those delivering treatment blind to treatment assignment?	☐	☐	☐	☐
6.	Were outcomes assessors blind to treatment assignment?	☐	☐	☐	☐
7.	Were treatment groups treated identically other than the intervention of interest?	☐	☐	☐	☐
8.	Was follow up complete and if not, were differences between groups in terms of their follow up adequately described and analyzed?	☐	☐	☐	☐
9.	Were participants analyzed in the groups to which they were randomized?	☐	☐	☐	☐
10.	Were outcomes measured in the same way for treatment groups?	☐	☐	☐	☐
11.	Were outcomes measured in a reliable way?	☐	☐	☐	☐
12.	Was appropriate statistical analysis used?	☐	☐	☐	☐
13.	Was the trial design appropriate, and any deviations from the standard RCT design (individual randomization, parallel groups) accounted for in the conduct and analysis of the trial?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion)

_____

^Reproduced from JBI (2014c)

Appendix H

JBI Critical Appraisal Checklist for Diagnostic Test Accuracy Studies^

JBI Critical Appraisal Checklist for Diagnostic Test Accuracy Studies

Reviewer_____Date_____

Author _____Year_____Record Number_____

	Yes	No	Unclear	Not applicable
1. Was a consecutive or random sample of patients enrolled?	☐	☐	☐	☐
2. Was a case control design avoided?	☐	☐	☐	☐
3. Did the study avoid inappropriate exclusions?	☐	☐	☐	☐
4. Were the index test results interpreted without knowledge of the results of the reference standard?	☐	☐	☐	☐
5. If a threshold was used, was it pre-specified?	☐	☐	☐	☐
6. Is the reference standard likely to correctly classify the target condition?	☐	☐	☐	☐
7. Were the reference standard results interpreted without knowledge of the results of the index test?	☐	☐	☐	☐
8. Was there an appropriate interval between index test and reference standard?	☐	☐	☐	☐
9. Did all patients receive the same reference standard?	☐	☐	☐	☐
10. Were all patients included in the analysis?	☐	☐	☐	☐

Overall appraisal: Include ☐ Exclude ☐ Seek further info ☐

Comments (Including reason for exclusion) _____

_____

^Reproduced from JBI (2014d)

Appendix I

JBI Levels of Evidence for Effectiveness^

LEVELS OF EVIDENCE FOR EFFECTIVENESS

Level 1 – Experimental Designs

Level 1.a – Systematic review of Randomized Controlled Trials (RCTs)

Level 1.b – Systematic review of RCTs and other study designs

Level 1.c – RCT

Level 1.d – Pseudo-RCTs

Level 2 – Quasi-experimental Designs

Level 2.a – Systematic review of quasi-experimental studies

Level 2.b – Systematic review of quasi-experimental and other lower study designs

Level 2.c – Quasi-experimental prospectively controlled study

Level 2.d – Pre-test – post-test or historic/retrospective control group study

Level 3 – Observational – Analytic Designs

Level 3.a – Systematic review of comparable cohort studies

Level 3.b – Systematic review of comparable cohort and other lower study designs

Level 3.c – Cohort study with control group

Level 3.d – Case – controlled study

Level 3.e – Observational study without a control group

Level 4 – Observational –Descriptive Studies

Level 4.a – Systematic review of descriptive studies

Level 4.b – Cross-sectional study

Level 4.c – Case series

Level 4.d – Case study

Level 5 – Expert Opinion and Bench Research

Level 5.a – Systematic review of expert opinion

Level 5.b – Expert consensus

Level 5.c – Bench research/ single expert opinion

^Reproduced from JBI (2014e)

Appendix J

JBI Levels of Evidence for Diagnosis^

LEVELS OF EVIDENCE FOR DIAGNOSIS

Level 1 – Studies of Test Accuracy among consecutive patients

Level 1.a – Systematic review of studies of test accuracy among consecutive patients

Level 1.b – Study of test accuracy among consecutive patients

Level 2 – Studies of Test Accuracy among non-consecutive patients

Level 2.a – Systematic review of studies of test accuracy among non-consecutive patients

Level 2.b – Study of test accuracy among non-consecutive patients

Level 3 – Diagnostic Case control studies

Level 3.a – Systematic review of diagnostic case control studies

Level 3.b – Diagnostic case-control study

Level 4 – Diagnostic yield studies

Level 4.a – Systematic review of diagnostic yield studies

Level 4.b – Individual diagnostic yield study

Level 5 – Expert Opinion and Bench Research

Level 5.a – Systematic review of expert opinion

Level 5.b – Expert consensus

Level 5.c – Bench research/ single expert opinion

^Reproduced from JBI (2014f)

Appendix K

Table K1

Table K1

Study Design, Sample Characteristics, Measures and Major Findings

Name	Title	Design	Country	Patient age	Parent	Diagnosis	Surgery Status	Severity	Sample size (group n)	Repeated Measures	Related Measures	Major related findings
Arafa et al. (2008)	Quality of life among parents of children with congenital heart disease	Quan	Egypt	Mixed	Mixed	Mixed	Mixed	Mixed	800 parents (CHD n=400, minor conditions n=400)	N	SF-36 (Arabic version)	PCCHD had poorer self-rated health-related quality of life for all subscales (except for bodily pain) than parents of children with minor conditions.
Berant et al. (2001)	Attachment style and mental health: A 1-year follow up study of mothers of infants with congenital heart disease	Quan	Israel	Neonates	Mothers	Mixed	Pre (at start)	Mixed	85 MCCHD	N	AAS; MHI; CAM; WCCL-SV. (Hebrew versions of all)	Mothers' attachment anxiety/avoidance following diagnosis was related to poor mental health. Attachment avoidance at diagnosis predicted deterioration of mental health over the next twelve months. Attachment style at diagnosis predicted maternal appraisal of, and coping with motherhood tasks. This was also related to mental health change, such that better mental health was found in those with more positive appraisal and a problem-focused approach.
Berant et al. (2003)	Marital satisfaction among mothers of infants with congenital heart disease: The contribution of illness severity, attachment style and the coping process	Quan	Israel	Neonates	Mothers	Mixed	Mixed	Mixed	85 MCCHD	Y (2 weeks post-birth, 1 year, 2 years)	ENRICH-SV; AAS; MHI; CAM; WCCL-SV. (Hebrew versions of all)	CHD severity and mothers' attachment anxiety/avoidance were related to reduced marital satisfaction. The negative effects of avoidance were increased by emotion-focused coping and the perception of motherhood as threatening.

Berant et al. (2008)	Mothers' attachment style, their mental health, and their children's emotional vulnerabilities: A 7-year study of children with congenital heart disease	Quan	Israel	Neonates	Mothers	Mixed	Mixed	Mixed	63 MCCHD	Y (2 weeks post-birth, 1 year, 7 years)	ENRICH-SV; AAS; MHI; CAM; WCCL-SV. (Hebrew versions of all)	Maternal attachment avoidance at diagnosis predicted deterioration in mental health and marital satisfaction over the next seven years, especially when the CHD was severe. Maternal attachment anxiety/avoidance at diagnosis was associated with emotional problems and poor self-image of children seven years later.
Bevilacqua et al. (2013)	Birth of a child with congenital heart disease: Emotional reactions of mothers and fathers according to time of diagnosis	Quan	Italy	Infants (surgery <4 months)	Mixed	Mixed	Required	Severe	72 PCCHD (n=38 couples)	N	GHQ-30); BDI-II; SF-36	60-82% of parents experienced psychological distress, and 20-46% experienced clinical levels of depression (mothers significantly higher than fathers). Timing of diagnosis had no significant effect on distress or depression for mothers/fathers.
Bratt et al. (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	Qual	Sweden	Neonates	Mixed	Mixed	Mixed	Mixed	12 PCCHD (n=6 couples)	N	Interview	Parents with a prenatally diagnosed foetus needed: counselling and support for decision making; continued support during pregnancy, support to plan for the near future.
Bright et al. (2013)	Infant cardiac surgery and the father-infant relationship: Feelings of strength, strain and caution	Mixed	Australia	Infants	Fathers	Mixed	Completed	Mixed	63 FCCHD	N	Interview; PPAS	Paternal postnatal attachment, patience and tolerance was at similar levels to community norms. Pleasure and interaction, and affection and pride, were significantly lower for CHD fathers. 37% of fathers reported a strong relationship with their infant. 17% indicated apprehension or condition-specific worry. Qualitative themes included: relationship strength (with infant); relationship strain; behaviours to promote strength in relationship; conscientiousness about health and the future; desire to maintain normalcy; respect and admiration; not enough interaction; medical condition facilitated the relationship.

Brosig, Mussato et al. (2007)	Psychosocial outcomes for preschool children and families after surgery for complex congenital heart disease	Quan	United States	Pre-schoolers (3-6 years)	Mixed	TGA, HLHS	Completed	Severe	26 PCCHD (TGA n=13; HLHS n=13)	N	PSI; PBC	Parents of children with HLHS reported more overall stress and greater negative impact on the family than parents of children with TGA. HLHS children were seen as having characteristics that made them difficult to parent. Parents of both groups of children were more permissive in their parenting style than parents of healthy controls. 96% of parents indicated that they were closer to their child because of the CHD experience.
Brosig, Whitstone et al. (2007)	Psychological distress in parents of children with severe congenital heart disease: The impact of prenatal versus postnatal diagnosis	Mixed	United States	Neonates /Infants	Mixed	Mixed	Mixed	Mixed	34 PCCHD (prenatally diagnosed n=10 couples; postnatally diagnosed n=7 couples)	Y (diagnosis, birth, six mth f/u)	BSI; Interview	At diagnosis, PCCHD had greater distress than normative samples regardless of the timing of diagnosis. Six months post-birth, the postnatal group scored within the normative range but the prenatal group was still more distressed. The more severe the CHD, the greater the distress.
Bruce & Sundin (2012)	Experience of support for parents of adolescents with heart defects - supported to be supportive	Qual	Sweden	Adoles-cents	Mixed	Mixed	Mixed	Mixed	5 PCCHD	N	Interview	PCCHD reported gaining fulfilment from actively attempting to support their children, their broader family, school staff, and other people involved with their children. The support given to others was influenced by support received from care providers.
Bruce et al. (2014)	Mothers' lived experiences of support when living with young children with congenital heart disease	Qual	Sweden	Mixed (3-12 years)	Mothers	Mixed	Mixed	Mixed	6 MCCHD	N	Interview	Mothers experienced support that ranged through good, poor and non-existent. Those that received person and family-centered support were more likely to cope better with the stresses of parenting a CHD child.
Bruce et al. (2016)	Support for fathers of children with heart defects	Qual	Sweden	Mixed (2-12 years)	Fathers	Mixed	Mixed	Mixed	10 FCCHD	N	Interview	Fathers identified feeling safe and supported when involved in the care of their child and/or when in a relationship with others. They identified negative experiences when support was not present.

Cantwell-Bartl & Tibballs (2015)	Psychosocial responses of parents to their infants' diagnosis of hypoplastic left heart syndrome	Qual	Australia	Infants	Mixed	HLHS	Post	Severe	29 PCCHD	N	Interview	PCCHD reported being devastated by the CHD diagnosis and having experienced loss and other stressors. 83% reported that diagnosis and the aftermath was the worst time of their lives. All parents chose surgery for their infant but differed on when they made this decision.
Carey et al. (2002)	Maternal factors related to parenting young children with congenital heart disease	Mixed	United States	Preschoolers (2-5 years)	Mothers	Mixed	Mixed	Moderate to Severe	60 mothers (CHD n=30; HC n=30)	N	Interview; PBC; PSI-SF; ECBI; BSQ	MCCHD and MHC were similar in their mother-child interactions and in their quantitative ratings of behaviour/stress. However qualitatively, MCCHD reported high levels of vigilance.
Clark & Miles (1999)	Conflicting responses: The experiences of fathers of infants diagnosed with severe congenital heart disease	Qual	United States	Neonates	Fathers	Mixed	Mixed	Severe	8 FCHD	Y (hospital and 12 month f/u)	Interview	FCCHD experienced conflicting reactions to the birth of their child: hiding intense emotions vs being strong for others; feeling loss of control vs needing to retain control; feeling fear about infant mortality/vulnerability vs becoming attached; feeling sadness and loss at CHD vs feeling joy in meeting the infant and becoming a father.
Connor et al. (2010)	The meaning of cost for families of children with congenital heart disease	Qual	United States	Mixed (0-5years)	Mixed	Mixed	Mixed	Mixed	20 PCCHD	N	Interview	PCCHD identified the cost of CHD in more than monetary terms. Uncertainty and life-style change was found across emotional, family burden and financial cost domains. Parent SES and severity of CHD were linked to higher levels of stress all domains. Prenatal diagnosis triggered early discussion and planning for financial uncertainty.
Davis et al. (1998)	Psychological adaptation and adjustment of mothers of children with congenital heart disease: Stress, coping and family functioning	Quan	United States	Mixed (9 days - 13.6 years)	Mothers	Mixed	Mixed	Mixed	52 MCCHD	N	Structured interview for stress appraisal; HSUP; MHLC; WCQ; FES; BSI	When CHD severity and maternal education was controlled for, a palliative/emotion-focused approach to coping (self-blame, avoidance and wishful thinking) and high levels of daily stress explained 38% of the variance in maternal adjustment.

Diffin et al. (2016)	Stress and distress in parents of neonates admitted to the neonatal intensive care unit for cardiac surgery	Quan	United Kingdom	Infants	Mixed	Mixed	Post	Mixed	211 parents (CHD n=69; HC n=142)	Y (prior to discharge, 6 mths, 12mths)	HADS); CISS; FSS; PSS- NICU	PCCHD had higher levels of depression and anxiety than controls. The NICU period had the highest scores for depression and anxiety, scores associated with the appearance/behaviour of the infant and the sights/sounds of NICU. Parents reported that parental role alteration in NICU was the most stressful part. Higher anxiety was associated with lower task-focused coping.
Doherty et al. (2009)	Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease	Quan	Ireland	Neonates	Mixed	Mixed	Mixed	Severe	Parent/s of 70 infants (n unclear)	N	BSI; COPE; MWS, SOS, FES	Mothers had higher rates of clinically elevated psychological distress than fathers, and used different coping styles to fathers. Distress in parents was predicted more by knowledge, coping style, family functioning and subjective worry, than by the presence of CHD or demographic factors.
Dulfer et al. (2015)	Parental mental-health moderates the efficacy of exercise training on health-related quality of life in adolescents with congenital heart disease	Quan	Netherlands	Mixed (10-16years)	Mixed	ToF, SV (Fontan circulation)	Post	Severe	112 parents/children CHD (intervention child n=34, parents n=34; control child n= 22, parents n=22)	Y (baseline pre-intervention and 2mth f/u)	GHQ-28) - Dutch version;	The mental health of PCCHD was comparable to or better than normative samples. Parental mental health (anxiety, insomnia and severe depression) moderated the impact that an exercise program had on HRQoL for adolescents with ToF (or Fontan circulation).
Fischer et al. (2012)	Caregiver anxiety upon discharge for neonates with congenital heart disease	Quan	United States	Neonates	Mixed	Mixed	Mixed	Mixed	59 PCCHD (of 38 children)	N	STAI	Five percent of PCCHD (neonates) admitted to NICU reported significant state anxiety (14% borderline and 81% no anxiety) just prior to discharge. Higher levels of education were associated with higher anxiety levels.

Franck et al. (2010)	Parents stress levels during children's hospital recovery after congenital heart surgery	Mixed	United Kingdom	Mixed (0-16 years)	Mixed	Mixed	Pre (at start)	Mixed	211 PCCHD (cohort 1 n=110; cohort 2 n=101)	Y (1 day preop; 3, 5, 8, 15 das post op if still in hospital)	PSS-CH; Structured interviews	PCCHD experienced moderate stress during hospitalisation regardless of CHD severity. Parents perception of child's CHD severity correlated with post-operative morbidity. Mothers and fathers had similar stress levels, however PCCHD born outside of the United Kingdom, those living in more vulnerable communities, and those with a child under a year old were at greater risk.
Franich-Ray et al. (2013)	Trauma reactions in mothers and fathers after their infant's cardiac surgery	Quan	Australia	Neonates	Mixed	Mixed	Post	Mixed	143 PCCHD (mothers n=77; Fathers n=55)	N	ASDS	33.8% MCCHD and 18.2% FCCHD met criteria for Acute Stress Disorder one month post-surgery. MCCHD had higher scores for all symptom clusters than fathers (except for dissociation). Dissociation was the most commonly reported symptoms by MCCHD and FCCHD (at clinical levels for 26% PCCHD). 83% of PCCHD had at least 1 symptom at a clinical level and only 11.4% had just 1.
Gardner et al. (1996)	Disturbed mother-infant interaction in association with congenital heart disease	Mixed	United Kingdom	Infants	Mothers	Mixed	Pre (at start)	Mixed	40 mothers (CHD n=20; HC n=20)	Y (2 days pre-surgery, 6 months post)	Filmed mother-infant interactions ; GHQ-30; Semi-structured interview	MCCHD showed less (and more variable) engagement and positive affect at pre-surgery and 6 month f/u than the control group. CHD severity was not related to either engagement or affect. MCCHD were significantly more distressed than the control group. Pre-surgery,75% had clinical levels of distress. At post-surgery no MCCHD met threshold. MCCHD had significantly higher positive affect at the 6 month f/u than at pre-surgery, however engagement remained the same.
Goldbeck & Melches (2005)	Quality of life in families of children with congenital heart disease	Quan	Germany	Mixed (7-20 years)	Mixed	Mixed	Mixed	Mix	138 parents/children CHD (parent/child dyads n=69)	N	ULQIE-29	Parent self-rated quality of life was significantly correlated with the child's quality of life as rated by the child and by the parent. Parents with lower self-rated quality of life were more likely to agree with child-rated quality of life than parents with high self-rated quality of life. Parent scores were generally in the higher level for quality of life ratings across all sub-scales, including psychological well-being.

Goldbeck & Melches (2006)	The impact of the severity of the disease and social disadvantage on quality of life in families with congenital cardiac disease	Quan	Germany	Mixed	Mixed	Mixed	Mixed	Mixed	132 PCCHD	N	ULQIE-29	Social disadvantage significantly impacted on the quality of life of PCCHD.
Gronning-Dale et al. (2012)	Mothers of infants with congenital heart defects: Well-being from pregnancy through the child's first six months	Quan	Norway	Neonates	Mothers	Mixed	Mixed	Mixed	61511 mothers (Mild CHD n=92; Moderate CHD n=50; Severe CHD n=70; Control n=61299)	Y (gestation week 30; 6 mths postpartum) .	SWLS; DES	MCCHD had similar satisfaction with life and feelings of joy as controls, and this remained unchanged from pregnancy through six months post-partum. At 6 months, mothers of children with severe CHD had higher feelings of anger than controls.
Gronning-Dale et al. (2013)	Well-being in mothers of children with congenital heart defects: A 3-year follow up	Quan	Norway	Neonates	Mothers	Mixed	Mixed	Mixed	44104 parents (mild CHD; n=79; moderate CHD n=36; severe CHD n=6); control n=43929)	Y (gestation week 30; 6 mths postpartum; 36 months postpartum) .	SWB	Mothers of children with severe CHD had significantly lower subjective well-being than controls at six months after birth, with a further decline by 36 months post-birth. Mothers of children with mild to moderate CHD did not differ from controls.
Gudmunds dottir et al. (1996)	Congenital heart defects and parent-adolescent coping	Qual	United States	Mixed (13-25)	Mixed	Mixed	Post	Mixed	16 parents/children CHD (parent/child dyads n=8)	N	Interview	Coping patterns of parent-adolescent dyads showed 1. similar coping (themes of: being accepting of each-other, mutual protection, normalising the illness, approaching the illness mechanically, keeping the illness in its place) and 2. dissimilar coping (themes of: withdrawing and problematising)

Harvey et al. (2013)	Experience of mothers of infants with congenital heart disease	Qual	United States	Infants	Mothers	Mixed	Pre (at start)	Severe	8 MCCHD	Y (journal entries prior to, during and after complex cardiac surgery)	Semi-structured interview	MCCHD identified an overarching theme of continuing to mother through everything, and six sub-themes: dealing with the unknown, navigating the medical world, intense fluctuating emotion, facing the possibility of infant death, and finding meaning and spiritual connection
Hearps et al. (2014)	Psychosocial risk in families of infants undergoing surgery for a serious congenital heart disease	Quan	Australia	Infants	Mixed	Mixed	Post	Mixed	39 PCCHD (of 29 children)	N	PAT (adapted for the study)	2.6% of PCCHD exhibited clinical psychosocial morbidity risk, 35.9% exhibited targeted risk and 61.5% had low risk. There were no differences based on time of diagnosis or CHD type (SVP vs BVP), however higher parental education predicted lower psychosocial risk.
Helfricht et al. (2009)	Psychometric evaluation and validation of the German version of the Aucte Stress Disorder Scale across two distinct trauma populations	Quan	Switzerland	Mixed (0-16years)	Mixed	Mixed	Post	Mixed	113 parent/adults (PCCHD n=61; Adult CHD patient(n=52)	Y (initial and 6 mth f/u)	ASDS - German Version	PCCHD had significantly higher scores on the ASDS than adult cardiac patients. 25 % of PCCHD were diagnosable as having Acute Stress Disorder (vs 4% of the adult cardiac patients). The German version of ASDS was found to be psychometrically sound.
Hoehn et al. (2004)	Parental decision making in congenital heart disease	Mixed	United States	Neonates	Mixed	Mixed	Pre (at start)	Mixed	52 PCCHD of 31 neonates (prenatal diagnosis n=29; postnatal diagnosis n=23)	N (quant pre-surgery, qual post-surgery)	STAI); LOT; LES; Semi-structured interview	At time of surgery, there were no differences in anxiety, life events or optimism for MCCHD based upon timing of diagnosis. FCCHD were more optimistic, had less negative life events and were less anxious if they had received a prenatal vs postnatal diagnosis. Thematic analysis of post-surgery interviews, found that PCCHD felt they had made a genuine choice regarding surgery (regardless of timing of diagnosis).

Jordan et al. (2014)	Early mother-infant relationships after cardiac surgery in infancy	Mixed	Australia	Infants	Mothers	Mixed	Post	Mixed	97 MCCHD	N	MPAS); EPDS); Semi-structured interview	MCCHD exhibited normal levels of attachment to their infant. Thematic analysis of interviews identified four themes related to the impact of CHD: anxiety and worry; caregiving behaviour; enhanced emotional ties; and bonding difficulties. Those identifying bonding difficulties had lower attachment scores and higher post-natal depression scores, and were more likely to have had a prenatal diagnosis. Lower attachment scores were associated with higher post-natal depression scores.
Kocyildirim et al. (2007)	Intra-operative imaging in paediatric cardiac surgery: The reactions of parents who requested and watched a video of the surgery performed on their child	Qual	United Kingdom	Neonates /Infants (15 days to 7 months)	Mixed	Mixed	Post	Mixed	17 PCCHD (of 10 infants)	N	Semi-structured interview	No PCCHD reported distress at watching a video of their child's cardiac surgery. Motivation for watching the video included a desire for learning and wanting to know what had happened during surgery. Post-viewing PCCHD did not change attitude towards the surgical teams. PCCHD used the videos to share the experience with family and friends.
Kosta et al. (2015)	Parental expereinces of their infant's hospitalization for cardiac surgery	Qual	Australia	Neonates	Mixed	Mixed	Post	Mixed	154 PCCHD	N	Structured interview	PCCHD identified a number of challenges: dealing with the illness, surgery and recovery; dealing with systemic barriers to staying near their baby; struggling to obtain daily necessities at the hospital; and balancing home and hospital life. Areas for improvement were also identified: increasing availability of resources/facilities; improving quality of information; and increasing emotional support. PCCHD found that their relationships with hospital staff was the most common source of support during admission.
Lan et al. (2007)	Maternal experiences making a decision about heart surgery for their young children with congenital heart disease	Qual	Taiwan	Preschoolers (3 years)	Mothers	Mixed	Post	Mixed	9 MCCHD	N	Interview	Factors involved in the maternal process of decision making for their CHD child in relation to surgery included: understanding the surgery in detail; role pressure; developing care-taking ability; maintaining family functioning whilst in preparation for surgery; and deliberate consideration to make the correct decision

Landolt (2011)	Predictors of parent quality of life after child open heart surgery: A 6 month prospective study	Quan	Switzerland	Mixed (0-16 years)	Mixed	Mixed	Post	Mixed	232 PCCHD	Y (Discharge from surgery, 6 mth f/u)	SF-36; PDS - German version; IFS	The psychological functioning of PCCHD was low following open-heart surgery but within six months had normalised. However, PCCHD who reported an increased impact of the child's illness on the family, were at higher risk of continued low mental health.
Lawoko & Soares (2002)	Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children	Quan	Sweden	Mixed (0-20 years)	Mixed	Mixed	Mixed	Mixed	1497 parents (CHD n=1092; HC n=293; OD n=112)	N	SCL-90-R; BHS	PCCHD were more distressed and had higher rates of hopelessness than PHC and PCOD, with MCCHD more distressed and with less hope than FCCHD, and MCCHD the most distressed and least hopeful of all groups. No differences were found between PHC and PCOD. A significant number of PCCHD experienced distress and hopelessness at levels equal to psychiatric outpatients and above that of a depressed sample. Economic variables explained more of the variance in hopelessness and distress than disease variables.
Lawoko & Soares (2003a)	Quality of life among parents of children with congenital heart disease, parents of children with other diseases and parents of healthy children	Quan	Sweden	Mixed (0-20 years)	Mixed	Mixed	Mixed	Mixed	1497 parents (CHD n=1092; HC n=293; OD n=112)	N	GQL-SV; ISSI; SCL-90-R BHS	Higher psychological distress was associated with lower quality of life. PCCHD reported lower quality of life and higher psychological distress than PHC. MCCHD had the lowest quality of life and experienced more psychological distress than MCCOD and MHC. Mothers experienced significantly more distress than fathers.
Lawoko & Soares (2003b)	Social support among parents of children with congenital heart disease, parents of children with other diseases and parents of healthy children	Quan	Sweden	Mixed (0-20 years)	Mixed	Mixed	Mixed	Mixed	632 PCHD	Y - 12 mths apart	ISSI; SCL-90-R; BHS	All parents reported low availability of social support (with no significant differences between PCCHD and PCOD and PHC). Mothers had lower availability of social support than fathers, and MCCHD were the lowest. Variation in the availability of social support was due more to care giving time, psychological distress, than it was due to gender, disease or severity.

Lawoko & Soares (2004)	Satisfaction with care: A study of parents of children with congenital heart disease and parents of children with other diseases	Quan	Sweden	Mixed (0-20 years)	Mixed	Mixed	Mixed	Mixed	1204 parents (CHD n=1092; OD n=112	N	SCL-90-R; BHS; ISSI, PPQ (modified for study); CSQ (modified for study)	PCCHD were more satisfied than PCOD with medical care and waiting periods for treatment. Fathers were more satisfied than mothers with the attitudes of staff, with MCCHD having the least satisfaction in this domain. Reduced satisfaction with care was associated more with having a younger child, unemployment, financial burden, psychological distress and social isolation, than it was with type of disease, severity or parent gender.
Lawoko & Soares (2006)	Psychosocial morbidity among parents of children with congenital heart disease: A prospective longitudinal study	Quan	Sweden	Mixed (0-20 years)	Mixed	Mixed	Mixed	Mixed	1497 parents (CHD n=1092; HC n=293; OD n=112)	Y (initial, twelve month f/u)	SCL-90-R); BHS; ISSI; PPQ (modified for study); CSQ (modified for study)	Many PCCHD experienced depression (18%), anxiety (16-18%), somatization (31-38%) and hopelessness (16%) during initial measurement and at twelve-month follow up. 7-22% reported psychosocial problems persisting over the twelve-month period. Mothers consistently reported more severe symptoms across domains than fathers. CHD severity was not significantly related to parental psychosocial morbidity over time, however caregiving burden, dissatisfaction with care, social isolation and financial difficulties were associated with risk of long-term psychosocial problems.
Lee et al. (2007)	Parenting stress in mothers of children with congenital heart disease	Quan	Korea	Mixed (0-9years)	Mothers	HLHS	Post	Severe	16 PCCHD of 9 children)	N (however, 2 interviews taken per parent as possible)	PPUS; PRQ; PSI-SF	Parenting stress in PCCHD was significantly related to the age of the child (school-age child); maternal education (lower level); increased ambiguity; lack of information and clarity; and to reduced social support. Increased social support combined with using the internet as a source of information explained 39.4% of the variance in reduced parenting stress (33% for social support alone).
Lee & Rempel (2011)	Parenting children with hypoplastic left heart syndrome	Qual	Canada	Neonates /Infants (2months - 5 years)	Mixed	Mixed	Mixed	Mixed	51 MCCHD	N	Semi-structured interview	Three themes emerged for PCCHD: optimistic appraisal; normalisation; and perception of child vulnerability. Overall, parents used normalisation as a strategy to balance marvelling at their child's survival with worrying about their child's vulnerability.

Levert et al. (2017)	Psychosocial needs of children undergoing an invasive procedure for a CHD and their parents	Quan	Netherlands	Mixed (0-18years)	Mixed	Mixed	Pre (at start)	Mixed	199 parents/children (PCCHD n=161, CCHD n=38)	N	Unvalidated questionnaire assessing care needs; LAS	Greater than 40% of PCCHD reporting needing increased psychosocial support. Those PCCHD with children aged 0-12 years had greater psychosocial need, than parents of adolescents.
Lok & Menahem (2004)	Parental perception of small ventricular septal defects in childhood	Qual	Australia	Mixed (1mth-16 years)	Mixed	VSD	Mixed	Mixed	40 PCCHD	N	Unvalidated questionnaire (open-ended)	PCCHD experienced distress and anxiety at the time of diagnosis
Majnemer et al. (2006)	Health and well-being of children with congenital cardiac malformations, and their families, following open-heart surgery	Quan	Canada	Infants	Mixed	Mixed	Post	Mixed	49 PCCHD	Y (infants developmental stage pre and post surgery, 12mths post-surgery and 18 mths post-surgery; parent measures at 5 year f/u)	PSI; CBCL; CHQ	Approximately 25% of PCCHD indicated high levels of stress, 20% had low levels of stress and 50% had stress within the normal range. Despite parents feeling that their child's quality of life was favourable 5 years after infant open-heart surgery, many felt stressed or defensive regarding their child (especially if behavioural difficulties were evident).
McCrossan et al. (2008)	Home support for children with complex congenital heart disease using videoconferencing via broadband: Initial results	Mixed	N. Ireland	Neonates (14-58 days)	Mixed	SV	Mixed	Severe	5 PCCHD	Y (pre-post conference call, multiple calls per participant)	STAI, interview	Parent STAI scores significantly reduced following in-home video-conferencing with a clinician in relation to concerns regarding their infant with complex CHD. Calls were qualitatively viewed as positive by clinicians and parents.

McCusker et al. (2010)	A controlled trial of early interventions to promote maternal adjustment and development in infants born with severe congenital heart disease	Quan	N. Ireland	Infants	Mothers	Mixed	Post	Mixed	54 MCCHD (intervention n=31; control n=23)	Y (pre and six month f/u)	STAI, MWS, COPE	At six-month follow-up, MCCHD receiving the CHIP intervention had reduced anxiety and worry and more positive appraisal of their situation.
McCusker et al. (2012)	A randomised controlled trial of interventions to promote adjustment in children with congenital heart disease	Quan	N. Ireland	Preschoolers (4-5 years)	Mothers	Mixed	Post	Mixed	90 MCCHD (intervention n=45; control n=45)	Y (pre and ten month f/u)	BSI; MWS; IFS	MCCHD in the CHIP-school intervention group had significantly less emotional problems, and experienced significantly less personal strain, in the period between intervention and f/u than the control group. They also had significant decreases in psychological distress at f/u (with controls showing an increase).
Meakins et al. (2015)	Parental vigilance in caring for their children with Hypoplastic Left Heart Syndrome	Qual	Canada	Mixed (2-60 months)	Mixed	HLHS	Post	Severe	41 PCCHD	N	Interview	Themes related to caring for a child with HLHS included a process of PCCHD comparing what was out of their hands and what was in their hands. In developing complex care skills, vigilance was common in PCCHD. PCCHD recognised that their vigilance was sometimes excessive, however was appropriate in some situations.
Odegard et al. (2002)	A survey of parental satisfaction during parent present induction of anaesthesia for children undergoing cardiovascular surgery	Mixed	United States	Mixed (3 months to 12 years)	Mixed	Mixed	During	Mixed	183 PCCHD	N	Unvalidated questionnaire	96.7% of PCCHD felt prepared for parent present induction (PPI) of their child's anaesthesia for heart surgery and that it was of benefit for both themselves and their child.

Pelchat et al. (1999)	Adaptation of parents in relation to their 6-month-old infants' type of disability	Quan	Canada	Infants (6 months)	Mixed	Mixed	Mixed	Mixed	144 parents of 72 children (CHD n=36; Downs Syndrome n=32; cleft lip/palette n=38; HC n=38)	N	SAM; PSI; QHS IDPSQ-14	PCCHD and parents of children with Downs Syndrome report higher levels of psychological distress and parenting stress than PHC or parents of those with cleft lip and/or palette. Mothers reported greater levels of stress than fathers consistently across groups.
Pinelli (1981)	A comparison of mothers concerns regarding the care-taking tasks of newborns with congenital heart disease before and after assuming their care	Qual	Canada	Neonates (0-2 months)	Mothers	Mixed	Mixed	Mixed	10 MCCHD	Y (interviews at discharge from hospital and one month post)	Structured interview	The number of concerns reported by mothers of infants being discharged from hospital increased pre to post discharge by 61%.
Pinto et al. (2016)	Modifiers of stress related to timing of diagnosis in parents of children with complex congenital heart disease	Quan	United States	Infants	Mixed	Mixed	Mixed	Mixed	202 PCCHD of 105 families (prenatal diagnosis n=60 families - mothers n=60, fathers n=59; postnatal diagnosis n=45 families - mothers n=45, fathers n=38)	Y (diagnosis, birth, 4-9 mth f/u)	BSI	PCCHD with a prenatal diagnosis were lower on anxiety and global stress at diagnosis and birth (but not at follow-up) than PCCHD with a postnatal diagnosis. In those with a prenatal diagnosis, mothers were more stressed than fathers.

Pridham et al. (2010)	Internal working models of parenting: motivations of parents of infants with a congenital heart defect	Qual	United States	Infants	Both	Mixed	Mixed	Severe	Parent/s of 25 CHD infants	Y (1-2, 4-6 ,12 mths)	Interview	All parents of infants with CHD identified being motivated to support their infant through care-taking and to promote their child's development/growth. All bar one parent set were motivated to guard/protect their own well-being, and this was most common at the 4-6 month interview. Most parents expressed a motivation to guard the infant's health at the 12 month interview.
Rahimianafar et al. (2015)	Anxiety determinants in mothers of children with congenital heart diseases undergoing cardiac surgery	Quan	Iran	Infants	Mothers	Mixed	Mixed	Mixed	69 MCCHD	N	STAI, unvalidated questionnaire	Maternal stress in MCCHD was significantly higher than that found in normative samples. Increased stress was found in younger mothers and those with younger infants. Differences in stress based on maternal education level, history of family CHD, infant hospitalisation history and family support were not significant.
Redshaw et al. (2011)	Narratives of the heart: telling the story of children with a cardiac condition through a bead program	Qual	Australia	Mixed (0-15 years)	Both	Mixed	Mixed	Mixed	14 parents/child of 11 families (MCCHD n=10; FCCHD n=1; child with CHD n= 3)	N	Interview	Exploration of a narrative intervention (cardiac bead program) revealed that PCCHD found the beads as important for: acknowledgement; connection with others; and imagining the future. The beads were seen as a tool for encouraging/uplifting and as symbolism. An overarching theme of telling the CHD child's story through the beads was also apparent.
Rempel & Harrison (2007)	safeguarding precarious survival: parenting children who have a life threatening heart disease	Qual	Canada	Mixed (2 months - 5years)	Both	HLHS	Post	Severe	16 PCCHD of 9 children (mothers n=9; fathers n=7)	Y (multiple interviews)	Unstructured interviews	Over-arching themes emerged of PCCHD protecting the child's precarious survival whilst safeguarding their own, and extreme parenting in the context of constant uncertainty.
Rempel et al. (2009)	Is "treat your child normally" helpful advice for parents of survivors of treatment for hypoplstic left heart syndrome?	Qual	Canada	Mixed (2 months - 5years)	Both	HLHS	Post	Severe	17 PCCHD of 9 children (mothers n=9; fathers n=7)	Y (multiple interviews)	Unstructured interview	Normalisation was used as a coping strategy to counter uncertainty by PCCHD. PCCHD set their own milestones and celebrations for their CHD children.

Rona et al. (1998)	Anxiety and depression in mothers related to severe malformation of the heart of the child and foetus	Quan	United Kingdom	Foetus/ Infant	Mothers	Mixed	Mixed	Severe	108 MCCHD (prenatal true positive diagnosis n=28; prenatal false positive diagnosis n=40; child with CHD n=40)	N	HADS	Medium term psychological well-being (anxiety and depression) was assessed 6-10 months after referral to fetal or paediatric cardiology units. Anxiety was higher in groups of women with a confirmed diagnosis (prenatal and postnatal) than in those with a false positive screen for CHD. Young females diagnosed prenatally had the highest anxiety. Depression was highest in those with a postnatal diagnosis than those diagnosed prenatally or given a false positive screen. Women who terminated pregnancies based on prenatal diagnosis were depressed for an extended period.
Rychik et al. (2013)	Maternal psychological stress after prenatal diagnosis of congenital heart disease	Quan	United States	Foetus (gestation 24-30 weeks)	Mothers	Mixed	Pre	Mixed	59 MCCHD	N	IES-R; STAI; BDI-II; COPE; DAS	39% of women with a prenatally diagnosed foetus had clinically significant traumatic stress; 22% had clinically significant depression, and 31% had clinically significant state anxiety. Higher anxiety and depression was significantly associated with reduced partner satisfaction. When income and partner satisfaction were controlled for, increased anxiety, depression and traumatic stress was significantly associated with denial.
Sarajuuri et al. (2012)	Patients with univentricular heart in early childhood: Parenting stress and child behaviour	Quan	Finland	Infants (17-23 months)	Mixed	HLHS, other UVH	Post	Severe	83 parents (HLHS n=23); other UVH n=14; HC (n=46)	N	PSI, CBCL	Mothers and fathers of children with HLHS were significantly more stressed than those of healthy controls. They also reported significantly more child behaviour concerns than controls. Parents of children with functioning UVH (non-HLHS) did not differ from healthy controls.
Sira et al. (2014)	coping strategies in mothers of children with heart defects - a closer look into spirituality and and internet utilisation	Quan	United States	Mixed (0-18+ years)	Mothers	Mixed	Mixed	Mixed	175 MCCHD	N	CHIP; SIBS; unvalidated open-ended questionnaire	The most frequently used coping pattern for MCCHD was communication with other PCCHD and understanding the medical situation. The least frequent was promoting self-esteem and psychological stability. Effective family functioning (integration and optimism) was contributed to by higher reliance upon spirituality. Better understanding of the medical situation was contributed to by more frequent use of the Internet.

Sklansky et al. (2002)	maternal psychological impact of fetal echocardiography	Quan	United States	Foetus/ Infants	Mothers	Mixed	Mixed	mixed	235 women with CHD child/foetus (CHD foetus n=213; CHD child n=22)	N	Unvalidated questionnaire	Fetal echocardiography that detected CHD was associated with higher maternal anxiety and reduced happiness with being pregnant. Normal fetal echocardiography was associated with decreased maternal anxiety, increased perception of closeness to the foetus and increased happiness. MCCHD who had had fetal echocardiography had better relationships with their partners and felt less responsible for the CHD.
Solberg et al. (2011a)	Emotional reactivity in infants with CHD and maternal symptoms of postnatal depression	Quan	Norway	Foetus	Mothers	Mixed	Mixed	mixed	242 MCCHD (severe n=73; MCCHD mild/moderate n=169)	Y (30 week gestation; 6 mth postpartum)	SCL-4 DEP; EPDS; ICQ	After controlling for infant emotional reactivity, mothers of infants with severe CHD had significantly elevated symptoms of postnatal depression as compared to those with mild/moderate CHD at 6 months post-partum.
Solberg et al. (2011b)	Long-term symptoms of depression and anxiety in mothers of infants with congenital heart defects	Quan	Norway	Foetus	Mothers	Mixed	Mixed	Mixed	44562 mothers (CHD n=162; cohort n=44400)	Y (30 week gestation, 6 and 18mth postpartum)	SCL-8	Mothers of infants with severe CHD showed significantly higher levels of anxiety and depression at 6 months and 18 months postpartum as compared to the overall cohort. Mothers of infants with mild and moderate CHD were no different to the overall cohort.
Solberg et al. (2012)	Trajectories of maternal mental health: a prospective study of mothers of infants with congenital heart disease	Quan	Norway	Foetus	Mothers	Mixed	Post	mixed	36578 mothers (CHD n=141; cohort n=36,437)	Y (30 week gestation; 6, 18 and 36 mths postpartum)	SCL-8	Mothers of children with severe CHD had significantly higher levels of anxiety and depression than the overall cohort at 6, 18 and 36 months postpartum.
Sparacino et al. (1997)	the dilemmas of parents of adolescents and young adults with congenital heart disease	Qual	United States	Mixed	Both	Mixed	Post	mixed	8 PCCHD	N	Interview	The following themes were identified by parents of adolescents with CHD: trying to cope; impact on the family; social isolation vs integration; strategies and dilemmas for CHD management; dilemmas of disclosure; normality dilemmas; and uncertainty.

Spijkerboer et al. (2007)	Long-term psychological distress and coping styles in parents of children and adolescents who unerwent invasive treatment for congenital heart disease	Quan	The Netherlands	Mixed (7-15 years)	Mixed	Mixed	Mixed	Mixed	583 parents (CHD n=109; reference group n=474)	N	GHQ; UCL	PCCHD tended to have lower levels of distress (somatic symptoms, sleeplessness, serious depression and anxiety) than the reference group. Fathers had significantly less somatic symptoms than mothers.
Svavarsdottir & McCubbin (1996)	parenthood transition for parents of an infant diagnosed with a congenital heart condition	Quan	United States	Infants (1 month to 1 year)	Mixed	Mixed	Mixed	Mixed	142 PCCHD (couples n= 71)	N	FPI; FILE; CMC; CHIP	There were differences in the way in which PCCHD spent caregiving time: mothers attended to physical needs and fathers attended to emotional/developmental needs of the infants. When the infant was younger, fathers reported higher caregiving demands but also more positive coping strategies (self, family, health care situation). No significant relationships between coping strategies and family/caregiving demands was found for mothers. First time parents had lower levels of family systems demands than parents with additional children.
Tak & McCubbin (2002)	Family stress, perceived social support and coping following the diagnosis of a child's congenital heart disease	Quan	United States	Mixed (under 12 years)	Mixed	Mixed	Mixed	mixed	Parents of 92 CHD children	N	FILE; PRQ; CHIP	PCCHD who reported higher levels of social support had lower levels of family stress and reported higher levels of positive coping. Child gender, current age, age at diagnosis and CHD severity were not related to stress. Younger parents reported more helpful coping styles, however mothers and fathers differed in type of style used.
Torowitz et al. (2010)	infant temperament and parental stress in 3-month old infants after surgery for complex congenital heart disease	Quan	United States	Neonates	Mothers	SV and BV	Post	mixed	258 mothers/infants (mother-infant dyads n=129)	N	PSI, EITQ	Stress for MCCHD (related to caregiving demand) was significantly higher than for MHC. For mothers of SVP infants, negative infant mood and difficulty in soothing the infant predicted increased total life stress and stress within the child domain. Mothers of SVP infants rated significantly higher stress than MHC and mothers of children with BVP in five of the six child domain subscales.

Utens et al. (2002)	Does age at the time of elective cardiac surgery or catheter intervention in children influence the longitudinal development of psychological distress and styles of coping of parents	Quan	Germany	Mixed	Mixed	Mixed	Pre at baseline	Mild-Moderate	634 women/men (CHD cardiac surgery mothers n=67; CHD cardiac surgery fathers n=60; CHD catheter mothers n=17, CHD catheter fathers (n=16); reference group women n=258; reference group men n=216)	Y (1 mth pre surgery and post surgery f/u at 18 mths)	GHQ-28; UCL	PCCHD who underwent cardiac surgery reported significantly higher psychological distress than PCCHD who underwent cardiac catheterisation. Distress decreased pre to post procedure for PCCHD (regardless of whether surgery or catheterisation was undertaken). Overall, child age at time of cardiac procedure did not influence parental distress.
Uzark & Jones (2003)	parenting stress and children with heart disease	Quan	United States	Mixed (2-12 years)	Mixed	Mixed	Mixed	Mixed	80 PCCHD (mothers n=70; fathers n=10)	N	PSI	PCCHD had higher levels of parenting stress than normative samples, particularly in relation to characteristics of the CHD child that made them difficult to parent. Clinically significant stress was found in approximately 30% of PCCHD. This stress was not related to CHD severity, SES or time since most recent surgery but was related to difficulties in disciplining/setting limits for the CHD child. The older the child the more stress experienced by the parents.

Vrijmoet-Wiersma et al. (2009)	A multicentric study of disease-related stress, and perceived vulnerability in parents of children with congenital heart disease	Quan	The Netherlands	Mixed (10 months - 8 years)	Mixed	Mixed	Post	Mixed	196 PCCHD of 131 children (mothers n=114; fathers n=82)	N	PIP - SF; GHQ; PSI-SF; STAI; CVS	There were no reported differences in parenting, generic or disease-related stress between PCCHD and reference groups. State anxiety was higher in MCCHD. MCCHD and FCCHD had significantly higher levels of perceived vulnerability than PHC. Risk factors for anxiety and perceived vulnerability included ethnicity, number of surgical procedures and time since last surgery. CHD severity did not predict parental stress levels, however parents of children with HLHS had higher levels of stress than other parents.
Werner et al. (2014)	The impact of an infant's severe CHD on the family	Quan	Switzerland	Infants	Mixed	Mixed	Post	Mixed	147 PCCHD of 104 infants	Y (6mths and 12 mths post-surgery)	IFS; F-Soz-U	No differences on IFS between MCCHD and FCCD. The impact on family was increased by reduced social support and the presence of a genetic disorder. PCCHD most frequently endorsed that they were living on a roller coaster and were not thinking of having any more children.
White et al. (2016)	A therapeutic recreation camp for children with congenital heart disease: examining impact on the psychosocial well-being of parents	Qual	Canada	Mixed (8-12 years)	Both	Mixed	Post	severe	9 PCCHD	Y (pre camp and post camp)	Interview	A camp for CCHD allowed parents to: let children grow; let children be independent; reduce their own overprotective and restricting behaviours; and receive respite care.
Wilson & Chando (2015)	Parental experiences with a hospital based bead programme for children with congenital heart disease	Quan	Australia	Mixed	Mixed	Mixed	Mixed	mixed	298 PCCHD of 166 children (mothers n=162); fathers n=136)	N	Survey designed for the study using previously published qualitative themes (Cronbach alpha for items .913)	A narrative therapy intervention (Heart Beads) was rated positively by PCCHD, although there was some concern regarding equity. 83% of MCCHD and 80% FCCHD reported that heart beads helped with understanding the child's CHD. 80% MCCHD and 58% FCHD felt that it improved CHD related communication. Fathers and mothers generally felt that their experience was acknowledged by hospital staff.

Wray & Sensky (2004)	Psychological functioning in paernts of children undergoing elective cardiac surgery	Quan	United Kingdom	Mixed (0 to 16.9 years)	Mixed	Mixed	Pre (at start)	Mixed	338 parents from 225 families (CHD n=102; bone marrow transplant n=114; PHC n=122)	Y (1day before and 1 year post surgery)	GHQ; DAS; UCL	Parents of children undergoing surgery for CHD and bone marrow transplant were significantly more distressed than those with healthy children. There were no differences between PCCHD who were cyanotic versus acyanotic. Post-treatment, parents were significantly less distressed.
Yildiz et al. (2009)	Distress levels in Turkish parents of children with congenital heart disease	Quan	Turkey	Mixed (3mth-12years)	Mixed	Mixed	Mixed	mixed	262 PCCHD	N	SCL-90-R – Turkish version	MCCHD had higher levels of overall distress, somatisation, anxiety and depression than FCCHD. The more severe the CHD, the greater the distress for both mothers and fathers.

Appendix L

Table L1

Table L1

Current Review Articles Cross-Referenced Against Other Reviews

Current Review	Existing Reviews		
	Soulvie et al. (2012)	Wei et al. (2015)	Jackson et al. (2015)
Arafa et al. (2008)		X	X
Berant et al. (2001)			
Berant et al. (2003)			
Berant et al. (2008)			X
Bevilacqua et al. (2013)			
Bratt et al. (2015)			
Bright et al. (2013)			
Brosig, Mussato et al. (2007)	X	X	X
Brosig, Whitstone et al. (2007)	X	X	X
Bruce & Sundin (2012)			X
Bruce et al. (2014)			
Bruce et al. (2016)			
Cantwell-Bartl & Tibballs (2015)			
Carey et al. (2002)	X	X	X
Clark & Miles (1999)	X		
Connor et al. (2010)			
Davis et al. (1998)			X
Diffin et al. (2016)			X
Doherty et al. (2009)			
Dulfer et al. (2015)	X	X	X
Fischer et al. (2012)			
Franck et al. (2010)		X	
Franich-Ray et al. (2013)		X	X
Gardner et al. (1996)			
Goldbeck & Melches (2005)	X		
Goldbeck & Melches (2006)		X	X
Gronning-Dale et al. (2012)		X	X
Gronning-Dale et al. (2013)			
Gudmundsdottir et al. (1996)			
Harvey et al. (2013)			
Hearps et al. (2014)		X	
Helfricht et al. (2009)		X	
Hoehn et al. (2004)	X	X	
Jordan et al. (2014)		X	
Kocyildirim et al. (2007)			
Kosta et al. (2015)			
Lan et al. (2007)	X	X	
Landolt (2011)		X	X
Lawoko & Soares (2002)		X	X
Lawoko & Soares (2003a)			
Lawoko & Soares (2003b)			
Lawoko & Soares (2004)			
Lawoko & Soares (2006)		X	X

Lee et al. (2007)			X	
Lee & Rempel (2011)				
Levert et al. (2017)				
Lok & Menahem (2004)				
Majnemer et al. (2006)	X		X	
McCrossan et al. (2008)				
McCusker et al. (2010)			X	
McCusker et al. (2012)			X	
Meakins et al. (2015)				
Odegard et al. (2002)				
Pelchat et al. (1999)	X			
Pinelli (1981)	X			
Pinto et al. (2016)			X	
Pridham et al. (2010)			X	
Rahimianafar et al. (2015)				
Redshaw et al. (2011)				
Rempel & Harrison (2007)			X	
Rempel et al. (2009)			X	
Rona et al. (1998)				
Rychik et al. (2013)				
Sarajuuri et al. (2012)				X
Sira et al. (2014)				
Sklansky et al. (2002)	X			
Solberg et al. (2011a)			X	
Solberg et al. (2011b)				
Solberg et al. (2012)				
Sparacino et al. (1997)				
Spijkerboer et al. (2007)			X	X
Svavarsdottir & McCubbin (1996)	X			
Tak & McCubbin (2002)			X	X
Torowitz et al. (2010)	X		X	
Utens et al. (2002)			X	
Uzark & Jones (2003)			X	
Vrijmoet-Wiersma et al. (2009)			X	X
Werner et al. (2014)			X	
White et al. (2016)				
Wilson & Chando (2015)				
Wray & Sensky (2004)			X	X
Yildiz et al. (2009)			X	

Appendix M

Table M1

Table M1

Frequency of Measures (with abbreviations)

Measure	Abbreviation	Frequency	Percentage
Acute Stress Disorder Scale	ASDS	2	2.44
Adult Attachment Style Scale	AAS	3	3.66
Beck Depression Inventory - Second Edition	BDI-II	2	2.44
Beck Hopelessness Scale	BHS	5	6.10
Behaviour Screening Questionnaire	BSQ	1	1.22
Brief Symptom Inventory	BSI	5	6.10
Care of My Child	CMC	1	1.22
Child Behaviour Checklist	CBCL	2	2.44
Child Health Questionnaire	CHQ	1	1.22
Child Vulnerability Scale	CVS	1	1.22
Client Satisfaction Questionnaire	CSQ	2	2.44
Cognitive Appraisal of Motherhood	CAM	3	3.66
Coping Health Inventory for Parents	CHIP	3	3.66
Coping Inventory for Stressful Situations	CISS	1	1.22
Differential Emotions Scale	DES	1	1.22
Dyadic Adjustment Scale	DAS	2	2.44
Early Infancy Temperament Questionnaire	EITQ	1	1.22
Edinburgh Postnatal Depression Scale	EPDS	2	2.44
Evaluating and Nurturing Relationship Issues Communication and Happiness Scales - Short Version	ENRICH-SV	2	2.33
Eveberg Child Behaviour Inventory	ECBI	1	1.22
Family Environment Scale	FES	2	2.44
Family Inventory of Life Events	FILE	2	2.44
Family Profile Inventory	FPI	1	1.22
Family Support Scale	FSS	1	1.22
General Health Questionnaire	GHQ	3	3.66
General Health Questionnaire - 28 item version	GHQ-28	2	2.44
General Health Questionnaire - 30 item version	GHQ-30	2	2.44
Goteborg Quality of Life Scale - Short Version	GQL-SV	1	1.22
Hassels and Uplifts Scale	HSUP	1	1.22
Health Survey - 36 item	SF-36	3	3.66
Hospital Anxiety and Depression Scale	HADS	2	2.44
Impact of Events Scale Revisited	IES-R	1	1.22
Impact on Family Scale	IFS	3	3.66
Impact on Family Scale – Revised	IFS-R	1	1.22
Infant Characteristic Questionnaire	ICQ	1	1.22
Life Experiences Survey	LES	1	1.22
Life Orientation Test	LOT	1	1.22

Linear Analogue Scale	LAS	1	1.22
Maternal Postnatal Attachment Scale	MPAS	1	1.22
Maternal Worry Scale	MWS	3	3.66
Mental Health Inventory	MHI	3	3.66
Multidimensional Coping Inventory	COPE	3	3.66
Multidimensional Health Locus of Control Scales	MHLC	1	1.22
Paediatric Inventory for Parents -Short Form	PIP-SF	1	1.22
Parent Behaviour Checklist	PBC	2	2.44
Parental Stressor Scale – Hospitalisation	PSS-CH	1	1.22
Parental Stressor Scale – NICU	PSS-NICU	2	2.44
Parenting Stress Index	PSI	6	7.32
Parenting Stress Index - Short Form	PSI-SF	3	3.66
Parents' Perception Uncertainty in Illness Scale	PPUS	1	1.22
Paternal Postnatal Attachment Scale	PPAS	1	1.22
Personal Resources Questionnaire	PRQ	2	2.44
Post-Traumatic Distress Scale	PDS	1	1.22
Psychological Distress Index of the Quebec Health Survey	QHS IDPSQ-14	1	1.22
Psychosocial Assessment Tool	PAT	1	1.22
Pyramid Patient Questionnaire	PPQ	2	2.44
Satisfaction with Life Scale	SWLS	1	1.22
Schedule for Social Interaction	ISSI	4	4.88
Significant Others Scale	SOS	1	1.22
Social Support Questionnaire F-Soz	F-Soz-U	1	1.22
Spielberger State Trait Anxiety Scale	STAI	7	8.54
Spiritual Insight and Behaviour Scale	SIBS	1	1.22
Stress Appraisal Measure	SAM	1	1.22
Subjective Well-Being Index	SWB	1	1.22
Symptom Checklist - 4 item Depression	SCL-4 Dep	1	1.22
Symptom Checklist - 8 item version	SCL - 8	2	2.44
Symptom Checklist Revisited	SCL-90-R	6	7.32
Ulm Quality of Life Inventory for Parents	ULQIE-29	2	2.44
Utrecht Coping List	UCL	3	3.66
Ways of Coping Checklist - Short Version	WCCL-SV	3	3.66
Ways of Coping Questionnaire	WCQ	1	1.22
Other Tools			
Unvalidated measure	-	6	7.32
Unvalidated measure (reliability tested in current study)	-	1	1.22
Filmed interactions	-	1	1.22
Interview	-	30	36.59