Parental Coping After Their Baby's Diagnosis of Congenital Heart Disease

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Statement of Originality

*This thesis contains no material which has been accepted for the award of any other degree or diploma in any university or other tertiary institution and, to the best of my knowledge and belief, contains no material previously published or written by another person, except where due reference has been made in the text. I give consent to this copy of my thesis, when deposited in the University Library**, being made available for loan and photocopying subject to the provisions of the Copyright Act 1968.*

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Acknowledgement of Collaboration

I hereby certify that the work embodied in this thesis has been done in collaboration with other researchers including Dr Nadine Kasparian, Associate Professor Gary Sholler, Associate Professor Edwin Kirk, Associate Professor Bryanne Barnett and Associate Professor David Winlaw and was carried out within the Sydney Children's Hospitals Network (Westmead and Randwick). Other researchers included:

Dr Nadine Kasparian, PhD, Head of Psychological care at the Heart Centre for Children, The Sydney Children's Hospitals Network (Westmead and Randwick) and Senior Reserach Fellow (Paediatrics) at the School of Women's and Children's Health, UNSW Medicine, The University of New South Wales, Randwick, New South Wales. Dr Kasparian is the principal investigator on this project and is responsible for all aspects of the study. She has acted as principal supervisor for this thesis.

Associate Professor Gary Sholler, MBBS (Hons I) FRACP, is Director of The Sydney Children's Hospitals Network Cardiac Service (Westmead and Randwick) and Associate Professor (Paediatrics) at Discipline of Paediatrics and Child Health, Sydney Medical School, The University of Sydney. Dr Sholler has overseen and provided critical support in the clinical aspects of complex congenital heart disease.

Dr Edwin Kirk, MB BS, FRACP, PhD, is Staff Specialist in Medical Genetics in Sydney Children's Hospital and Conjoint Lecturer in Paediatrics at the University of New South Wales. Dr Kirk played a crucial role in the development of the principal research project and has overseen all components of the project ranging from recruitment to manuscript preparation.

Professor Bryanne Barnett, MB ChB, FRANZCP, MD, AM, is an expert in the field of perinatal and infant mental health. She is currently the Director of Clinical

Services and Chairman of the Board of Directors at Karitane in Sydney South West Area Health Services, as well as Conjoint Professor of Psychiatry at the University of New South Wales. Professor Barnett played an integral role in the development of the project and the interview guides utilised with parents as well as assisting in the preparation of manuscripts for publication.

Associate Professor David Winlaw, MB BS (Hons), MD, FRACS, is Head of Kids Heart Research at The Children's Hospital Westmead. Dr Winlaw has provided invaluable input regarding the surgical aspects of the project.

I have included as part of the thesis a statement clearly outlining the extent of collaboration, with whom and under what auspices.

Signed: Date:

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Structured Abstract

Scope and Purpose

Receiving a diagnosis of complex congenital heart disease (CHD) for one's baby is a highly stressful and potentially traumatic experience for parents and has the potential to have significant and long-lasting psychological effects (Rychik et al., 2013). Due to recent technological advancements it is now possible for mothers and fathers to receive a diagnosis during pregnancy (fetal[1] diagnosis) or postpartum (postnatal diagnosis). Currently, the literature is limited in the exploration of the similarities and differences in psychological outcomes and experiences amongst parents as a result of time of diagnosis and gender. Further, little is known about the coping strategies employed by mothers and fathers as they attempt to cope with such a diagnosis. Consequently, the present study had three main aims. First, to determine whether the stress and coping model proposed by Lazarus and Folkman (1984) can be applied to gain an understanding of parental coping in response to receiving their baby's diagnosis of complex CHD. Second, to explore thematic similarities and differences in the coping strategies employed by parents who received a fetal diagnosis with parents who received a postnatal diagnosis. Third, to explore thematic similarities and differences in the coping strategies utilised by mothers compared with fathers.

Methodology

A cross-sectional, mixed methods study design was utilised and data was taken from a larger study, previously conducted. Participants included mothers and fathers of a baby diagnosed with complex CHD during pregnancy (fetal diagnosis) or within

[1] This spelling of 'fetal' has been utilised throughout this thesis; as opposed to 'foetal', as it is has been deemed to be the more accurate term in the literature (Boyd & Hamilton, 1967).

the first six months of life (postnatal diagnosis). Participants were identified through the cardiology databases of the Sydney Children' Hospital Network, including the Children's Hospital at Westmead and the Sydney Children's Hospital, Randwick. In this sub-study, parents who completed their interview after the birth of their baby and before their infant reached 6 months of age were included.

Participants were first required to complete a semi-structured interview with Dr Nadine Kasparian, which was audio-recorded and transcribed verbatim. Two weeks after completing the interview, participants were then asked to complete a brief self-report questionnaire, from which, this study accessed demographic and medical information. Transcripts were coded utilising a modified analytic induction approach with the assistance of the qualitative data analysis software, *QSR NVivo10*.

Results

In total, twenty-five parents of fifteen babies completed twenty-two semi-structured interviews, with three couples choosing to complete the interview together. The sample consisted of six mothers and six fathers who received a fetal diagnosis and nine mothers and four fathers who received a postnatal diagnosis. After applying Lazarus and Folkman's model of stress and coping (1984), each of the three types of coping proposed (problem-focused, emotion-focused, meaning-focused) were evident within the narratives of participants. One main theme identified that could not be categorised within the model was parental pride and focus on the baby.

In the exploration of differences based on time of diagnosis it was found that parents who received a fetal diagnosis differed from parents who received a postnatal diagnosis in their level of preparedness, the nature and quality of distancing, meaning-focused coping and sources of reassurance. In comparing mothers and

fathers, differences arose in the use of confrontive coping, as mothers more frequently reported fighting to be close to their baby, and in the application of emotion-focused coping, as mothers more frequently reported using emotion-focused coping such as drawing upon social support. A larger proportion of mothers than fathers were also found to engage in meaning-focused coping, finding the benefit in the stressful situation that was their baby's condition.

General Conclusions and Implications

Parental coping with a diagnosis of complex CHD in their baby can be broadly categorised by the model of stress and coping proposed by Lazarus and Folkman (1984) with the exception of parental pride and focus on the baby. This finding suggests that while the model may be beneficial in guiding clinicians' generally in understanding the coping strategies employed by parents within this setting, it is limited in its ability to capture the importance of the relationship between parent and baby in coping with the difficulties resulting from a diagnosis.

The identified differences in coping strategies between diagnostic groups highlights the need for medical staff to help parents to feel better prepared for the birth and treatment of their baby. This will likely involve assisting parents in gathering information as problem-focused coping was identified as an important strategy employed by both mothers and fathers. Medical staff also have a role to play in helping parents to strengthen their attachment with their baby and find ways to separate their baby from their diagnosis of complex CHD. As a result of these findings, recommendations for further research include the use of quantitative measures, larger sample sizes and targeted exploration of the various forms of meaning-focused coping, emotion-focused coping and sources of reassurance utilised by parents.

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Critical Literature Review: Parents' experiences of fetal or early postnatal diagnosis of congenital abnormality: A systematic literature review

Abstract

Receiving a diagnosis of congenital anomaly in your baby can be a highly stressful and potentially traumatic experience for mothers and fathers. This systematic review was conducted with two aims in mind. First, to identify if the time a parent receives a diagnosis of a congenital anomaly in their baby has a significant impact upon their psychological response and psychosocial needs (fetal or postnatal). Second, to determine if psychological response and psychosocial needs vary between mothers and fathers.

A total of fifteen studies were identified via a systematic search of electronic databases containing international peer-reviewed journals from January 1806 to June 2014. Results across the fifteen studies were inconsistent. Five of the nine studies measuring the short-term impact of a diagnosis reported significant differences as a result of timing of diagnosis although two reported improved psychological outcomes for the postnatal diagnostic group and three improved psychological outcomes for the fetal diagnostic group. Six months after diagnosis, one study reported better outcomes for parents who received a postnatal diagnosis while two studies reported no differences as a result of timing of diagnosis. Only two studies were found that considered the long term impact of diagnosis, one study reported better outcomes after receiving a postnatal diagnosis while one study found no differences between the two groups.

In total, nine of the identified articles explicitly compared the experiences of mothers with fathers, with seven out of nine reporting mothers experienced greater

psychological distress and stronger emotional response. These findings emphasise a significant gap in the literature particularly due to the conflicting results presented.

Keywords: Fetal diagnosis, postnatal diagnosis, congenital anomalies, psychological stress, coping, parents, infant.

Introduction

It is estimated that, overall, congenital anomalies occur in approximately 6% of births worldwide, accounting for 6.7% of neonatal deaths (Christianson, Howson & Modell, 2006; World Health Organization [WHO], 2008). In Australia, 3.1% of all births have at least one congenital anomaly (Abeywardana & Sullivan, 2008), compared to 3% of births in the United States (Parker et al., 2010). Congenital anomalies, also referred to as birth defects in the literature, are defined as "structural or functional abnormalities, including metabolic disorders, which are present from birth" (WHO, 2010, p.1).

Three of the more common congenital disorders that have serious implications for the health and development of infants include congenital heart disease (CHD), Down syndrome, and neural tube defects (WHO, 2010). CHD is the most common single organ abnormality in infants with 9.1 cases reported in every 1,000 births in Australia (NSW Ministry of Health, 2012), and 8.0 cases reported in every 1,000 births in Europe (Dolk, Loane & Garne, 2011). Down syndrome and neural tube defects also have a significant impact with 11.1 and 22.0 cases of Down syndrome reported per 10,000 births in Australia (Abeywardana & Sullivan, 2008) and Europe respectively (Loane et al., 2013) and 4.2 and 5 cases of neural tube defects per 10,000 births reported in Australia (Abeywardana & Sullivan, 2008) and the United States respectively (Wallingford, Niswander, Shaw & Finnell, 2013).

In the past, parents had predominantly learned about their baby's congenital anomaly after birth (postnatal diagnosis); however, with technological developments and more frequent antenatal screening has come an increase in the detection of congenital anomalies in the fetus (fetal diagnosis) (Boyd, Rounding, Chamberlain, Wellesley & Kurinczuk, 2012). Across an 18 year period in Europe, fetal diagnosis

of congenital anomalies rose from 48% of cases in 1991-1993 to 61% of cases in 2006 -2008 (Boyd et al., 2012). In Australia, at the Royal Children's Hospital in Melbourne, 14.1% of complex CHD cases were diagnosed antenatally in 1994, compared to 39.7% of cases in 2003 (Chew, Stone, Donath & Penny, 2006).

From a medical perspective, fetal diagnosis is advantageous in comparison to postnatal diagnosis as it provides parents with ample opportunity to learn more about their baby's condition, to consider the treatment options available, potentially make alterations to pregnancy and birth management, to make detailed plans for intervention, to seek counselling, and to give consideration to terminating the pregnancy (Chew et al., 2006; Rychik et al., 2013). As an example, between 2002 and 2004 in Europe, 68% of cases of Down syndrome and 88% of neural tube defects were detected antenatally as a result of prenatal screening (Boyd et al., 2008). Of these detected cases, 88% of pregnancies were terminated (Boyd et al., 2008).

Receiving such a diagnosis can be a highly stressful and potentially traumatic experience for parents and families, and as a result, can have resounding psychological implications (Menahem & Grimwade, 2005; Howland, 2007; Rychik et al., 2013). It has been reported that receiving a diagnosis of congenital anomaly during pregnancy is experienced as a traumatic event by up to 88% of mothers and 83% of fathers (Aite et al., 2011). The ensuing treatment for the newborn can also give rise to serious psychological consequences. Studies indicate that, irrespective of their baby's condition, both mothers and fathers of a hospitalised neonate report significantly greater anxiety and depression in the postpartum period than parents of healthy neonates (Kong et al., 2013). Shaw et al (2006) reported 28% of mothers with infants in the neonatal intensive care unit (NICU) meet criteria of acute stress disorder and this symptomatology was associated with an inability to completely

fulfil aspects of the perceived parental role, such as help, hold and protect their baby. In contrast, family cohesion and expressiveness acted as a protective factor for parents and were associated with less psychological distress (Shaw et al., 2006). Specifically considering the example of CHD, Lawoko and Soares (2002) found parents of children with CHD were at greater risk of distress and hopelessness than parents of children with other diseases and parents of healthy children.

Thus, the aims of the present systematic review were twofold. First, to review the literature pertaining to parents who received either a fetal or postnatal diagnosis of congenital anomaly in their baby in order to determine if the timing of diagnosis influences the psychological responses and psychosocial needs of parents. Second, to determine if psychological responses and psychosocial needs vary when comparing mothers and fathers.

Methods

Literature search strategy and study inclusion criteria

The review was guided by the Preferred Reporting Items for Systematic Reviews and Meta-Analyses (PRISMA) guidelines (Moher, Liberati, Tetzlaff, & Altman 2009). Multiple strategies were utilised when conducting the search. As a starting point, electronic databases Medline, Medline In-Process, PsycINFO, and EMBASE were searched from January 1806 to June 2014 for eligible studies and review articles. Search terms utilised identified the two patient populations (fetal and postnatal diagnosis) and the target group of interest (parents and caregivers). When identifying fetal, postnatal and parent/caregiver groups, typical search terms included *foetal, fetal, fetus, prenatal, antenatal, postnatal, perinatal, neonatal, mother, father, parent, caregiver* and *family*. Limitations were placed on the search that meant all articles found were written in the English language and focused only on humans.

The abstracts of the identified articles were utilised to screen out ineligible studies, including articles that outlined epidemiological studies, molecular or biological studies, treatment trials, case reports, and clinical guidelines and recommendations, as well as articles that only considered child outcomes, did not include assessment of psychological factors, or did not compare a fetal and postnatal group. Reference lists of all eligible studies were hand searched to identify any relevant articles that were not revealed via database searches. Prolific authors in the field were also identified and individually searched for further publications.

Results

Study Selection

In total, 1,309 articles were identified using this search method. As stated above, the abstracts of these articles were read and screened utilising the above inclusion and exclusion criteria. Fifteen studies were identified that compared the psychosocial responses of parents who received their baby's diagnosis during pregnancy (fetal diagnosis) or postpartum (postnatal diagnosis) (see Table 1). The review incorporated all possible diagnoses of congenital anomaly with six of the 15 studies considering a variety of congenital anomalies/malformations and nine studies focusing on one specific congenital disorder, including; CHD (n=4), orofacial clefts (n=4) and Down syndrome (n=1). Nine of the 15 studies used quantitative methodologies, three used qualitative methodologies, and three utilised a mixed methods approach. The majority of studies identified were cross-sectional (10/15), whilst five studies utilised a longitudinal study design.

A relatively high proportion of the identified studies (9/15) included both mothers and fathers, with all but one of these studies explicitly comparing responses between the two groups. Five studies referred to "parents" but did not specify the

proportion of mothers and fathers, and only one study did not include fathers at all. Eight studies focused on parents of infants less than six months old, four studies included parents of children ranging in age from one to nine years, and three studies did not report the age of the child at time of study participation.

Mothers' and Fathers' psychological responses from birth to three months postpartum

In total, nine studies investigated the short-term impact of timing of diagnosis on mothers and fathers. Of these studies, five reported significant differences between parents who received a fetal, compared to a postnatal, diagnosis (Hoehn et al., 2004; Skari et al., 2006; Williams, et al., 2008; Fonseca, Nazare & Canavarro, 2012; Fonseca, Nazare & Canavaro, 2013). Moreover, seven out of eight studies reported significant differences in psychological outcomes between mothers and fathers (Hoehn et al., 2004; Skari et al., 2006; Skreden et al., 2010; Fonseca et al., 2012; Fonseca et al., 2013; Bevilacqua et al., 2013; Fonseca, Nazare & Canavarro, 2014).

Two studies reported lower psychological distress in parents who received a postnatal diagnosis (Skari et al., 2006; Fonseca et al., 2013). For example, Skari et al., (2006) found that mothers who received a fetal diagnosis reported the greatest psychological distress, both at time of birth (0-7 days after birth) and 6 weeks postpartum. In this study, parents who received a fetal diagnosis experienced higher psychological distress, anxiety, and depression than those who received a postnatal diagnosis (Skari et al., 2006). Psychological distress in parents was highest if the fetal diagnosis was made between weeks 25 and 30 of the pregnancy (Skari et al., 2006), and this finding remained consistent across the two time points (Skari et al., 2006).

In a more recent study exploring the emotional responses of parents one month after receiving their baby's diagnosis, Fonseca et al (2013) reported no differences in the category of emotions experienced by parents as a result of timing of diagnosis or gender. Findings did indicate, however, differences in the intensity of the emotions experienced, with mothers experiencing more intense guilt than fathers overall, and mothers who received a fetal diagnosis reporting greater sadness and anger than mothers who received a postnatal diagnosis (Fonseca et al., 2013).

In contrast, three studies found better understanding of the diagnosis, greater quality of life, less anxiety, and higher optimism amongst parents after receiving a fetal (as opposed to postnatal), diagnosis one month after birth (Williams et al., 2008; Fonseca et al., 2012; Hoehn et al., 2004). Parents who received a fetal diagnosis were found to have a greater understanding of CHD compared to parents in the postnatal diagnostic group; however, parents who received a fetal diagnosis were also reportedly four times more likely to report worry about taking their baby home after discharge (Williams et al., 2008).

When compared to parents of healthy infants, mothers and fathers of a baby with a congenital anomaly reported greater psychological distress, but equivalent quality of life (Fonseca et al., 2012). It was further found that mothers who received a fetal diagnosis demonstrated higher quality of life than their postnatal counterparts (Fonseca et al., 2012). Fonseca et al (2012) also reported differences associated with gender, with mothers reporting significantly higher levels of depression and anxiety, and lower levels of physical and psychological quality of life, than fathers.

Hoehn et al. (2004) investigated the psychological experiences of parents during their infant's hospital admission for cardiac surgery. While this study found no differences amongst mothers according to time of diagnosis, fathers who had

received a fetal diagnosis reported less anxiety, more optimism, and fewer negative life events compared to fathers who received a postnatal diagnosis (Hoehn et al., 2004). In interviews undertaken one week after their infant's surgery, mothers who received a fetal diagnosis discussed a change from initial feelings of grief and mourning to actively making plans, whilst mothers who had received a postnatal diagnosis discussed juggling an array of emotions including stress, whilst still trying to advocate for their newborn (Hoehn et al., 2004). Fathers who had received a fetal diagnosis raised concerns surrounding the financial pressures generated by the diagnosis, whilst fathers who had received a postnatal diagnosis were focused on getting the intervention required for their newborn and gathering as much information as possible (Hoehn et al., 2004). All parents reported feeling like they had made a genuinely informed choice for their baby to undergo surgery, regardless of the timing of diagnosis, and all reported contentment in their decision (Hoehn et al., 2004).

Four studies reported no differences in the short-term psychological responses of parents who received a fetal or postnatal diagnosis (Brosig, Whitstone, Frommelt, Frisbee & Leuthner, 2007; Bevilacqua et al., 2013; Fonseca et al., 2014; Nelson Goff et al., 2013). At time of birth and six weeks postpartum, Brosig et al., (2007) found no significant difference in psychological distress between mothers and fathers associated with time of diagnosis, although over 70% of parents in each group reported levels of emotional distress in the clinical range. Bevilacqua et al., (2013) reported similar findings, with no differences in reported levels of stress or depression between the fetal and postnatal diagnostic groups within the first three months postpartum. This study did, however, find a significantly higher proportion of mothers fell within the clinical range for depression (46%) and stress (82%) when

compared to fathers (20% depression, 61% stress) (Bevilacqua et al., 2013). A recent study by Fonseca et al (2014) also found no differences in reported psychological distress or quality of life one month after receiving either a fetal or postnatal diagnosis; however, mothers did report experiencing greater anxiety and depression, and lower physical quality of life, than fathers. In a qualitative study, Nelson Goff et al., (2013) retrospectively explored the experiences of parents after receiving their baby's diagnosis of Down syndrome. Parents were asked to recall their experiences several years after receiving the initial diagnosis, with the average age of the child at time of assessment being 4.84 years and 7.60 years in the fetal and postnatal groups, respectively (Nelson Goff et al., 2013). This study found parents experienced a gamut of emotions after receiving a diagnosis of Down syndrome, irrespective of the timing of diagnosis (Nelson Goff et al., 2013). Emotions included fear, grief and mourning, denial, guilt, and anger, with many parents describing that they had experienced all these emotions at the one time (Nelson Goff et al., 2013). So while four studies reported no differences between parents as a result of timing of diagnosis, two of the four studies reported significantly higher psychological distress in mothers (Brosig et al., 2007; Bevilacqua et al., 2013; Fonseca et al., 2014; Nelson-Goff et al., 2013).

Mothers' and fathers' psychological responses at six months after birth

Three studies identified in the review adopted a longitudinal design and examined the impact of a fetal or postnatal diagnosis six months after the birth (Skari et al. 2006; Brosig et al., 2007; Fonseca et al., 2014). Inconsistent with short-term psychological outcomes, Brosig et al. (2007) found that at six months parents who received a fetal diagnosis reported significantly greater psychological distress at six months postpartum. These data indicated that at the time of diagnosis both the fetal

and postnatal groups reported elevated levels of psychological distress, yet six months postpartum these levels had decreased in the postnatal diagnostic group whilst remaining consistently high in the fetal diagnostic group (Brosig et al., 2007). Consistent with the first two assessment points, no differences were reported between mothers and fathers in this study (Brosig et al., 2007).

The remaining two studies found no differences between parents due to time of diagnosis at six months postpartum (Skari et al., 2006; Fonseca et al., 2014). It was found that mothers mothers reported significantly higher anxiety than fathers at six months (Skari et al., 2006). Similarly, Fonseca et al (2014) found mothers reported significantly higher anxiety and lower psychological distress than fathers but reporting equivalent levels of depression and physical QoL.

Longer-term psychological responses amongst parents after fetal or postnatal diagnosis

Two studies were identified that considered the longer-term psychological and emotional consequences of timing of diagnosis for parents. Hunfeld et al. (1999) assessed psychological outcomes for mothers and fathers 12 months postpartum. They found mothers who received a fetal diagnosis reported higher total burden, stronger social impact, and greater grief and despair, as well as more difficulties with coping compared to mothers who received a postnatal diagnosis (Hunfeld et al., 1999). No differences were found amongst fathers in overall burden and grief according to time of diagnosis, although there was a trend for fathers who received a fetal diagnosis to report greater financial burden (Hunfeld et al., 1999). The study also found that mothers reported more personal strain than fathers; however mothers and fathers did not differ in overall burden or grief (Hunfeld et al., 1999). In further analyses it was found that levels of overall burden and grief in mothers were

correlated with levels of overall burden and grief reported by their partners (Hunfeld et al., 1999). In a nine-year longitudinal study, Skreden et al. (2010) found a sizable proportion of parents reported high levels of psychological distress (30%) and anxiety (28%), nine years after receiving their baby's diagnosis (Skreden et al., 2010). No differences were found amongst participants who had received a fetal versus postnatal diagnosis; however, mothers were found to experience significantly greater overall psychological distress and intrusive stress than fathers (Skreden et al., 2010).

It is important to highlight the period under assessment in each of the two studies is considerably different, with Hunfeld et al. (1999) considering one year after birth and Skreden et al. (2010) assessing outcomes nine years after diagnosis. It is therefore difficult to compare and contrast these conflicting results, which in turn, highlights a significant gap in the literature.

Impact of the severity of diagnosis

Three studies explored the impact of the severity of the diagnosis on the emotional responses of parents. Hunfeld et al. (1999) found the nature and severity of the diagnosis was correlated with the personal strain reported by parents. One year after birth, parents of babies who had received a complex diagnosis or multiple diagnoses reported greater burden compared to parents of infants with less complex conditions (Hunfeld et al., 1999). Brosig et al. (2007) found parents of children with complex CHD (e.g., hypoplastic left heart syndrome) were more likely to report clinically-significant levels of psychological distress than parents of children with less complex lesions (e.g., tetralogy of Fallot) (81% for complex, compared to 33% for less complex, CHD). Comparably, Fonseca et al (2013) found the type of congenital anomaly was associated with the intensity of emotions reported by

mothers (sadness, guilt and anger) but not fathers. For instance, a diagnosis of urinary system anomaly elicited greater reports of anger in mothers than a diagnosis of CHD, as well as greater guilt compared to mothers of a baby with a visible malformation (Fonseca et al., 2013).

Psychosocial needs identified by parents who received a fetal or postnatal diagnosis for their offspring

Berggren, Hansson, Uvemark, Svensson and Becker (2012) investigated the psychosocial needs and emotional responses of parents after receiving either a fetal or postnatal diagnosis of cleft lip with or without cleft palate. All parents recruited in the study received a consultation with a nursing assistant and possibly also a plastic surgeon during which time. This consultation was conducted with parents who received a fetal diagnosis prior to the birth of their baby while parents who received a postnatal diagnosis were provided a consultation within 48 hours after birth (Berggren et al., 2012). No differences were identified between the fetal and postnatal diagnostic groups in their rating of the information about cleft lip and counselling received (Berggren et al., 2012). Parents also did not differ in the emotions they reported after counselling (Berggren et al., 2012).

In a qualitative study, 85% of parents who received a fetal diagnosis reported feeling that the timing of diagnosis had helped them to better prepare psychologically for the birth of their baby and his or her treatment and 89% expressed gratitude that they received the diagnosis antenatally (Davalbhakta & Hall, 2000). Almost all parents (24/27 or 91%) who received a fetal diagnosis felt they had sufficient information, understood their baby's diagnosis and felt prepared, compared with 71% (45/63) of parents who received a postnatal diagnosis (Davalbhakta & Hall, 2000).

Also, in a qualitative exploration of parents' experiences, Nusbaum et al, (2008) found strong similarities in themes discussed by parents, regardless of time of diagnosis, including: shock after initially receiving the diagnosis, cause and information, support and disability. In addition, parents who received a fetal diagnosis proposed four unique themes: coping, preparation, disadvantages of fetal diagnosis, and alternative perspectives (Nusbaum et al., 2008). When parents in the fetal diagnostic group were asked to discuss the disadvantages of a fetal diagnosis, they were unable to identify any, instead speculating how difficult it would be to receive the diagnosis after their baby's birth (Nusbaum et al., 2008).

Impact of timing of diagnosis on maternal satisfaction with clinical care

Of the fifteen studies identified, one study focused solely on the experience of mothers. Robbins et al., (2010) found timing of diagnosis did not affect mothers' satisfaction with the information and support provided by medical staff. It was found, however, that mothers in the fetal diagnostic group perceived medical staff as more effective in assisting with the initiation of breastfeeding than mothers in the postnatal diagnostic group (Robbins et al, 2010).

Discussion

After undertaking a systematic search of the published literature, 15 studies were identified that examined the potential differences in parents' psychological experiences and needs after receiving either a fetal or postnatal diagnosis of congenital abnormality for their baby. Eight of these studies also directly compared the psychological experiences of mothers to those of fathers, thereby also taking potential gender differences into account.

Overall, results were mixed. Of the 15 identified studies, nine (60%) reported differences in psychological outcomes between parents who received a fetal or

postnatal diagnosis at least once across a range of assessment time points. Inconsistent results were found across three different time points regarding the impact of timing of diagnosis. Of the nine studies exploring the short-term impact of the time of diagnosis, two studies indicated better psychological outcomes for parents who received a postnatal diagnosis (Skari et al., 2006; Fonseca et al., 2013), three indicated a fetal diagnosis was associated with lesser distress for parents (Williams et al., 2008; Fonseca et al., 2012; Hoehn et al., 2004) while a further four studies found no significant differences amongst parents as a result of time of diagnosis (Brosig et al., 2007; Bevilacqua et al., 2013; Fonseca et al., 2014; Nelson-Goff et al., 2013). At six months postpartum, two studies indicated no differences in psychological distress between diagnostic groups (Fonseca et al., 2014; Skari et al., 2006) and one study identified greater psychological distress in parents who received a fetal diagnosis (Brosig et al., 2007). Of the two studies that explored the long term impact of receiving a diagnosis one found significantly poorer psychological outcomes for mothers who received a fetal diagnosis (Hunfeld et al., 1999) while the other found no longer-term differences in psychological distress associated with timing of diagnosis (Skreden et al., 2010).

Currently, inconsistencies throughout the literature make it difficult to confirm or deny the hypothesised emotional advantages of a fetal diagnosis. In fact, in the current review, four studies reported parents who received a fetal diagnosis experienced greater psychological distress while three studies reported greater psychological distress experienced by the postnatal diagnostic group. Several explanations for these findings are plausible. Skari et al. (2006) suggest that such results indicate a fetal diagnosis acts as an ever-present psychological stressor for

parents, particularly if, as was the case with all participants in their sample, the congenital anomaly cannot be treated until after birth.

These results may also be indicative of the difficulties experienced by parents in terms of accessing health and psychological services in the antenatal period. Interestingly, the number of antenatal consultations a parent is able to attend after receiving a fetal diagnosis and before giving birth is correlated with parental anxiety, with parents who attend two or more consultations reporting lower anxiety levels at the time their baby is born (Aite et al., 2003). Specifically, parents have reported lower levels of anxiety after accessing antenatal counselling from specialist staff such as a paediatric surgeon or a neonatal nurse (Kemp, Davenport & Pernet, 1998). Parents may experience difficulties in accessing services for several reasons including; lack of services in their area, cost, transport, time restraints, personal beliefs, attitudes, cultural or religious beliefs or stigma surrounding mental health.

Clinicians working with parents may also unknowingly act as barriers to accessing services if they are unaware of services available or of the referral process, if they are unable to start a conversation with parents regarding the benefits of accessing services and the options available and if they are unable to recognise a parent who is experiencing distress.

In this review a wide range of diagnoses were considered, ranging from CHD to orofacial clefts to Down syndrome. It is theorised that the nature of the malformation, the implications of the diagnosis and the corresponding stigma may also play a role in the level of psychological distress experienced by parents. For example, some diagnoses are physically visible while others are not, some have significant long-term implications while some diagnoses have multiple treatments and options available compared to others that have very few alternatives available.

Further, various stigma associated with different diagnoses may elicit varying levels of psychological distress in parents. In a 2011 study, Lee and Rempel highlighted normalization as an important coping strategy employed by parents of children with hypoplastic left heart syndrome as they try to come to terms with the diagnosis. This strategy involves viewing and treating their child as a normal child, pushing against the possibility for their child to be defined by their diagnosis (Lee & Rempel, 2011). It is hypothesised that the use of normalization would be more effective with some diagnoses compared to others, for example, with infants with CHD compared to infants with Down syndrome. Consequently, variations between diagnostic groups and between mothers and fathers may be a result of variations in diagnoses rather than differences due to timing of diagnosis or gender.

Perhaps the most significant discrepancy identified was recognised via longitudinal analyses. As stated above, Skari et al. (2006) found differences in psychological distress between diagnostic groups one week after their baby's birth were not sustained six months postpartum. In contrast, Brosig et al. (2007) reported no differences amongst parents in psychological outcomes at the time of birth but found parents who learned of the congenital anomaly during pregnancy experienced greater distress six months after birth than those who had received the diagnosis after birth. In this study, parents in both groups experienced high levels of psychological distress at the time of birth; however, only parents in the postnatal group reported lower levels of distress six months after birth (Brosig et al., 2007). The authors suggest this may be due to more complex CHD diagnoses for babies in the fetal group compared to those in the postnatal group but other possible explanations also need to be considered (Brosig et al., 2007). The differences between the two studies may also be due to differences in the sample populations utilised as Skari et al.

(2006) included babies with congenital anomalies while Brosig et al. (2007) looked specifically at CHD. Another possible factor is the emotional consequences of receiving a diagnosis of congenital anomaly in your baby during pregnancy. Previously, mothers have reported after receiving a fetal diagnosis in their baby, the pregnancy ceases to be a pleasurable experience and attention is focused only on the baby (Catlin, Askelsdottir, Conroy & Rempel, 2008). Parents who receive a fetal, as opposed to a postnatal, diagnosis are not given the opportunity to meet and bond with their newborn before learning of their condition. These parents may be left instead with a sense of loss and disappointment due to missing out on a normal pregnancy and childbirth, a feeling that has also been expressed by parents of preterm babies (Jackson, Ternestedt & Schollin, 2003). It is speculated then that a fetal diagnosis, if not appropriately addressed in the antenatal period, has the potential to have lasting psychological and emotional consequences for parents.

Mixed results were also found when comparing the psychological outcomes for mothers and fathers; with the majority of studies reporting mothers experienced greater psychological distress than fathers. One possible explanation as to why mothers reported greater psychological distress is due to differences in appraisal. Previously it has been reported that women appraise stressful situations as more stressful than males (Eaton & Bradley, 2008), which may lead to differences in response to the situation and consequently possible differences in psychological outcomes. Cultural beliefs and gender stereotypes may also be at play with fathers reportedly less inclined to accurately report their experiences and responses than mothers (Mirowsky & Ross, 1995). A further possible explanation is that mothers are likely to experience greater distress than fathers during pregnancy and birth, even when the baby is healthy. Skari et al. (2002), in a study of parents of healthy

newborns, found overall mothers reported greater distress, anxiety, social dysfunction and intrusive stress when compared to fathers. These differences did, however, decrease six weeks after birth, with mothers and fathers reporting symptoms of depression typically seen in the general population by six weeks postpartum (Skari et al., 2002).

The present review is also helpful in identifying several important limitations in the literature, including the relatively narrow number of studies examining the topic, small sample sizes (Hunfeld et al., 1999; Brosig et al., 2007; Williams et al., 2008; Fonseca et al., 2012; Bevilacqua et al., 2013; Fonseca et al., 2013; Fonseca et al., 2014), use of non-validated measures of psychological stress (Davalbhakta & Hall, 2000; Williams et al., 2008; Berggren et al., 2012; Fonseca et al., 2013) retrospective study design (Berggren et al., 2012; Fonseca et al., 2013), and a lack of diversity in the study samples (e.g., higher proportions of mothers than fathers, as well as greater representation in the postnatal, compared to fetal, diagnostic groups) (Davalbhakta & Hall, 2000; Robbins et al., 2010; Nelson Goff et al., 2013). Also of concern, a lack of consistency in the psychological measures and psychometric tools utilised (see Table 2). Moreover, nine of the 15 samples utilised were heterogeneous in that they included a range of different diagnoses, of varying severity and complexity, and did not analyse outcomes for each of the different diagnoses (Hunfeld et al., 1999; Hoehn et al., 2004; Skari et al., 2006; Brosig et al., 2007; Skreden et al., 2010; Berggren et al., 2012; Fonseca et al., 2012; Nelson Goff et al., 2013; Fonseca et al., 2014). Studies that investigated “congenital anomalies” inherently encounter this limitation as they include a range of diagnoses whilst other studies also encountered this limitation due to variances in the severity or complexity of the diagnosis included. Finally, all studies are limited by selection bias, as none of

the studies in the review invited parents who elected to terminate their pregnancy after receiving a fetal diagnosis to participate in the study. Similarly, several studies recognised the possibility that parents who had received severe diagnoses may have elected not to participate thereby biasing the sample (Skari et al., 2006; Brosig et al., 2007). For instance, in Brosig et al (2007) the response rate varied greatly between the fetal (91% response rate) and postnatal diagnostic groups (44% response rate). A proportion of parents who decline to participate (89%) in the postnatal diagnostic group had received a diagnosis for their child that was classified as severe (Brosig et al., 2007).

Implications for Clinical Practice

The current literature does not satisfactorily lend support for better psychological and emotional outcomes in parents who receive either a fetal or a postnatal diagnosis. As a consequence, the theoretical psychological and emotional benefits of receiving a fetal diagnosis have yet to be substantiated. Due to this current conjecture in the literature regarding fetal versus postnatal diagnosis, it is not currently possible to determine what would be considered best practice based simply on the psychological and emotional outcomes of the parents of children with a congenital anomaly.

The review does report that fathers have better psychological and emotional outcomes when compared to mothers. The strong differences identified between mothers and fathers also cause consideration for clinicians to offer varying levels of support to mothers and fathers in order to best meet their individual needs.

Further, findings highlight the importance of access to health and psychological services for parents, in both the antenatal and postnatal periods. It is also suggested that clinicians working with these parents are given assistance in offering and

accessing services on their behalf, particularly looking at breaking down gender stereotypes and mental health stigma in order to help parents to engage with offered services. Findings further suggest clinicians can play a role in helping parents in their appraisal of the diagnosis and understanding the implications for themselves and for their baby.

Implications for Research

The discrepancies identified in this review give credence to an argument for further research into this particular area, particularly due to the increasing likelihood of a fetal diagnosis for many conditions (Boyd et al., 2012). Specifically, further research is required in order to determine if a fetal or a postnatal diagnosis is more effective in helping to achieve better psychological and emotional outcomes for parents.

This review has particularly highlighted the need for further exploration of the long-term impact of a diagnosis of congenital anomaly on parents. Brosig et al's (2007) findings call into question the potential lasting effects a fetal diagnosis may cause and as a consequence highlights the need for better access to services in the antenatal period.

The review identified a need for greater consistency within the literature in the psychological constructs measured and the corresponding psychometric tools utilised in order to generate results that are comparative across studies. It further highlighted the importance of bearing in mind the impact of the nature, complexity, severity and corresponding stigma of the congenital anomaly diagnosed on the reported psychological distress experienced by parents.

Conclusions

Due to the conflicting results identified in the literature it is currently difficult to draw conclusions regarding the impact of timing of diagnosis nor gender on parents who receive a diagnosis of congenital anomaly in their baby. Further research is required to gain understanding into the short and long term psychological outcomes of parents after receiving such a diagnosis in order to better recognise and address their needs.

References

Abeywardana, S. & Sullivan, E. A. 2008. Congenital anomalies in Australia 2002–2003. Birth anomalies series no. 3 Cat. no. PER 41. Sydney: AIHW National Perinatal Statistics Unit.

AIHW 2011. Cardiovascular disease: Australian facts 2011. Cardiovascular disease series no. 35. Cat. no. CVD 53. Canberra: AIHW. Viewed 18 May 2014 http://www.aihw.gov.au/publication-detail/?id=10737418510.

Aite, L., Trucchi, A., Nahom, A., Zaccara, A., La Sala, E., & Bagolan, P. (2003). Antenatal diagnosis of surgically correctable anomalies: Effects of repeated consultations on parental anxiety. *Journal of Perinatology*, *23*(8), 652-654. doi:10.1038/sj.jp.7210992

Aite, L., Zaccara, A., Mirante, N., Nahom, A., Trucchi, A., Capolupo, I., & Bagolan, P. (2011). Antenatal diagnosis of congenital anomaly: A really traumatic experience? *Journal of Perinatology, 31*, 760- 763. doi:10.1038/jp.2011.22

Asplin, N., Wessel, H., Marions, L., & Georgsson Öhman, S. (2012). Pregnant women's experiences, needs, and preferences regarding information about malformations detected by ultrasound scan. *Sexual & Reproductive Healthcare*, *3*(2), 73-78. doi: 10.1016/j.srhc.2011.12.002

Berggren, H., Hansson, E., Uvemark, A., Svensson, H., & Becker, M. (2012). Prenatal compared with postnatal cleft diagnosis: What do the parents think? *Journal of Plastic Surgery and Hand Surgery*, *46*(3-4), 235-241. doi:10.3109/2000656X.2012.698416

Bevilacqua, F., Palatta, S., Mirante, N., Cuttini, M., Seganti, G., Dotta, A., & Piersigilli, F. (2013). Birth of a child with congenital heart disease: emotional reactions of mothers and fathers according to time of diagnosis. *The*

Journal of Maternal-Fetal & Neonatal Medicine, *26*(12), 1249-1253. doi:10.3109/14767058.2013.776536

Biehle, S. N., & Mickelson, K. D. (2011). Preparing for parenthood: How feelings of responsibility and efficacy impact expectant parents. *Journal of Social and Personal Relationships*, *28*(5), 668-683. doi: 10.1177/0265407510385493

Blue, G. M., Kirk, E. P., Sholler, G. F., Harvey, R. P., & Winlaw, D. S. (2012). Congenital heart disease: Current knowledge about causes and inheritance. *Medical Journal of Australia*, *197*(3), 155-159.

Boyd, P. A., DeVigan, C., Khoshnood, B., Loane, M., Garne, E., & Dolk, H. (2008). Survey of prenatal screening policies in Europe for structural malformations and chromosome anomalies, and their impact on detection and termination rates for neural tube defects and Down's syndrome. *BJOG: An International Journal of Obstetrics & Gynaecology*, *115*(6), 689-696. doi: 10.1111/j.1471-0528.2008.01700.x

Boyd, J. D., & Hamilton, W. J. (1967). Foetus—or Fetus? *British Medical Journal*, *1*(5537), 425.

Boyd, P. A., Rounding, C., Chamberlain, P., Wellesley, D., & Kurinczuk, J. J. (2012). The evolution of prenatal screening and diagnosis and its impact on an unselected population over an 18‐year period. *BJOG: An International Journal of Obstetrics & Gynaecology*, *119*(9), 1131-1140. doi: 10.1111/j.1471-0528.2012.03373.x

Brosig, C. L., Whitstone, B. N., Frommelt, M. A., Frisbee, S. J., & Leuthner, S. R. (2007). Psychological distress in parents of children with severe congenital heart disease: the impact of prenatal versus postnatal diagnosis. *Journal of Perinatology*, *27*(11), 687-692. doi:10.1038/sj.jp.7211807

Catlin, A., Áskelsdóttir, B., Conroy, S., & Rempel, G. (2008). From diagnosis to birth: Parents' experience when expecting a child with congenital anomaly. *Advances in Neonatal Care*, *8*(6), 348-354. doi: 10.1097/01.ANC.0000342768.94734.23

Centre for Epidemiology and Evidence. New South Wales Mothers and Babies 2010. Sydney: NSW Ministry of Health, 2012.

Chew, C., Stone, S., Donath, S. M., & Penny, D. J. (2006). Impact of antenatal screening on the presentation of infants with congenital heart disease to a cardiology unit. *Journal of Paediatrics and Child Health*, *42*(11), 704-708. doi: 10.1111/j.1440-1754.2006.00955.x

Christianson, A., Howson, C. P., & Modell, B. (2006). March of Dimes. *Global report on birth defects. The hidden toll of dying and disable children.* New York: White Plains, 29-35.

Davalbhakta, A., & Hall, P. (2000). The impact of antenatal diagnosis on the effectiveness and timing of counselling for cleft lip and palate. *British Journal of Plastic Surgery*, *53*(4), 298-301. doi:10.1054/bjps.2000.3330

Dolk, H., Loane, M., & Garne, E. (2010). The prevalence of congenital anomalies in Europe. In *Rare Diseases Epidemiology* (pp. 349-364). Springer Netherlands.

Fonseca, A., Nazaré, B., & Canavarro, M. C. (2011). Patterns of parental emotional reactions after a pre-or postnatal diagnosis of a congenital anomaly. *Journal of Reproductive and Infant Psychology*, *29*(4), 320-333. doi:10.1080/02646838.2011.634398

Fonseca, A., Nazaré, B., & Canavarro, M. C. (2012). Parental psychological distress and quality of life after a prenatal or postnatal diagnosis of congenital anomaly:

A controlled comparison study with parents of healthy infants. *Disability and Health Journal*, *5*(2), 67-74. doi:10.1016/j.dhjo.2011.11.001

Fonseca, A., Nazaré, B., & Canavarro, M. C. (2013). Clinical determinants of parents' emotional reactions to the disclosure of a diagnosis of congenital anomaly. *Journal of Obstetric, Gynecologic, & Neonatal Nursing*, *42*(2), 178-190. doi: 10.1111/1552-6909.12010

Fonseca, A., Nazaré, B., & Canavarro, M. C. (2014). Parenting an infant with a congenital anomaly An exploratory study on patterns of adjustment from diagnosis to six months post birth. *Journal of Child Health Care*, *18*(2), 111-122. doi: 10.1177/1367493512473856

Gupta, N., Leven, L., Stewart, M., Cheung, M., & Patel, N. (2014). Transport of infants with congenital heart disease: benefits of antenatal diagnosis. *European Journal of Pediatrics*, *173*(5), 655-660. doi: 10.1007/s00431-013-2231-0

Hoehn, K. S., Wernovsky, G., Rychik, J., Tian, Z. Y., Donaghue, D., Alderfer, M. A., ... & Nelson, R. M. (2004). Parental decision-making in congenital heart disease. *Cardiology in the Young*, *14*(03), 309-314. doi: http://dx.doi.org/10.1017/S1047951104003099

Howland, L. C. (2007). Preterm birth: implications for family stress and coping. *Newborn and Infant Nursing Reviews*, *7*(1), 14-19. doi:10.1053/j.nainr.2006.12.008

Hunfeld, J. A., Tempels, A., Passchier, J., Hazebroek, F. W., & Tibboel, D. (1999). Brief report: Parental burden and grief one year after the birth of a child with a congenital anomaly. *Journal of Pediatric Psychology*, *24*(6), 515-520. doi: 10.1093/jpepsy/24.6.515

Jackson, K., Ternestedt, B. M., & Schollin, J. (2003). From alienation to familiarity:

experiences of mothers and fathers of preterm infants. *Journal of Advanced Nursing*, *43*(2), 120-129. doi: 10.1046/j.1365-2648.2003.02686.x

Kemp, J., Davenport, M., & Pernet, A. (1998). Antenatally diagnosed surgical anomalies: The psychological effect of parental antenatal counseling. *Journal of Pediatric Surgery*, *33*(9), 1376-1379. doi:10.1016/S0022-3468(98)90011-2

Kong, L. P., Cui, Y., Qiu, Y. F., Han, S. P., Yu, Z. B., & Guo, X. R. (2013). Anxiety and depression in parents of sick neonates: A hospital‐based study. *Journal of Clinical Nursing*, *22*(7-8), 1163-1172. doi: 10.1111/jocn.12090

Lawoko, S., & Soares, J. J. (2002). Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children. *Journal of Psychosomatic Research*, *52*(4), 193-208. doi:10.1016/S0022-3999(02)00301-X

Lee, A., & Rempel, G. R. (2011). Parenting children with hypoplastic left heart syndrome: Finding a balance. *Journal for Specialists in Pediatric Nursing*, *16*(3), 179-189. doi: 10.1111/j.1744-6155.2011.00289.x

Loane, M., Morris, J. K., Addor, M. C., Arriola, L., Budd, J., Doray, B., ... & Dolk, H. (2013). Twenty-year trends in the prevalence of Down syndrome and other trisomies in Europe: Impact of maternal age and prenatal screening. *European Journal of Human Genetics*, *21*(1), 27-33. doi:10.1038/ejhg.2012.94

Mathers, C., Fat, D. M., & Boerma, J. T. (2008). *The global burden of disease: 2004 update*. World Health Organization.

Menahem, S., & Grimwade, J. (2005). Pre-natal counselling—helping couples make decisions following the diagnosis of severe heart disease. *Early Human Development*, *81*(7), 601-607. doi:10.1016/j.earlhumdev.2005.02.001

Moher, D., Liberati, A., Tetzlaff, J., & Altman, D. G. (2009). Preferred reporting

items for systematic reviews and meta-analyses: the PRISMA statement. *Annals of Internal Medicine*, *151*(4), 264-269. doi:10.7326/0003-4819-151-4-200908180-00135

Nelson Goff, B. S., Springer, N., Foote, L. C., Frantz, C., Peak, M., Tracy, C., ... & Cross, K. A. (2013). Receiving the Initial Down Syndrome Diagnosis: A Comparison of Prenatal and Postnatal Parent Group Experiences. *Mental Retardation*, *51*(6), 446-457. doi: http://dx.doi.org/10.1352/1934-9556-51.6.446

Nusbaum, R., Grubs, R. E., Losee, J. E., Weidman, C., Ford, M. D., & Marazita, M. L. (2008). A qualitative description of receiving a diagnosis of clefting in the prenatal or postnatal period. *Journal of Genetic Counseling*, *17*(4), 336-350. doi: 10.1007/s10897-008-9152-5

Parker, S. E., Mai, C. T., Canfield, M. A., Rickard, R., Wang, Y., Meyer, R. E., ... & Correa, A. (2010). Updated national birth prevalence estimates for selected birth defects in the United States, 2004–2006. *Birth Defects Research Part A: Clinical and Molecular Teratology*, *88*(12), 1008-1016. doi: 10.1002/bdra.20735

Poh, H. L., Koh, S. S. L., & He, H. G. (2014). An integrative review of fathers' experiences during pregnancy and childbirth. *International Nursing Review*, *61*(4), 543-554. doi: 10.1111/inr.12137

Robbins, J. M., Damiano, P., Druschel, C. M., Hobbs, C. A., Romitti, P. A., Austin, A. A., ... & Burnett, W. (2010). Prenatal diagnosis of orofacial clefts: association with maternal satisfaction, team care, and treatment outcomes. *The Cleft Palate-Craniofacial Journal*, *47*(5), 476-481. doi: 10.1597/08-177

Rychik, J., Donaghue, D. D., Levy, S., Fajardo, C., Combs, J., Zhang, X., ... &

Diamond, G. S. (2013). Maternal psychological stress after prenatal diagnosis of congenital heart disease. *The Journal of Pediatrics*, *162*(2), 302-307. doi: 10.1016/j.jpeds.2012.07.023

Shaw, R. J., Deblois, T., Ikuta, L., Ginzburg, K., Fleisher, B., & Koopman, C. (2006). Acute stress disorder among parents of infants in the neonatal intensive care nursery. *Psychosomatics*, *47*(3), 206-212.

Sholler, G. F., Kasparian, N. A., Pye, V. E., Cole, A. D., & Winlaw, D. S. (2011). Fetal and post‐natal diagnosis of major congenital heart disease: Implications for medical and psychological care in the current era. *Journal of Paediatrics and Child Health*, *47*(10), 717-722. doi: 10.1111/j.1440-1754.2011.02039.x.

Sivarajan, V., Penny, D. J., Filan, P., Brizard, C., & Shekerdemian, L. S. (2009). Impact of antenatal diagnosis of hypoplastic left heart syndrome on the clinical presentation and surgical outcomes: The Australian experience. *Journal of Paediatrics and Child Health*, *45*(3), 112-117. doi: 10.1111/j.1440-1754.2008.01438.x.

Skari, H., Malt, U. F., Bjornland, K., Egeland, T., Haugen, G., Skreden, M., ... & Emblem, R. (2006). Prenatal diagnosis of congenital malformations and parental psychological distress—a prospective longitudinal cohort study. *Prenatal Diagnosis*, *26*(11), 1001-1009.doi**:** 10.1002/pd.1542

Skari, H., Skreden, M., Malt, U. F., Dalholt, M., Ostensen, A. B., Egeland, T., & Emblem, R. (2002). Comparative levels of psychological distress, stress symptoms, depression and anxiety after childbirth—a prospective population‐based study of mothers and fathers. *BJOG: An International Journal of Obstetrics & Gynaecology*, *109*(10), 1154-1163. doi: 10.1111/j.1471-0528.2002.00468.x

Skreden, M., Skari, H., Malt, U. F., Haugen, G., Pripp, A. H., Faugli, A., & Emblem, R. (2010). Long‐term parental psychological distress among parents of children with a malformation—A prospective longitudinal study. *American Journal of Medical Genetics Part A*, *152*(9), 2193-2202. doi: 10.1002/ajmg.a.33605

Springett, A, & Morris J, K. (2012). Congenital Anomaly Statistics 2010: England and Wales. London: British Isles Network of Congenital Anomaly Registers.

van der Bom, T., Zomer, A. C., Zwinderman, A. H., Meijboom, F. J., Bouma, B. J., & Mulder, B. J. (2011). The changing epidemiology of congenital heart disease. *Nature Reviews Cardiology*, *8*(1), 50-60. doi:10.1038/nrcardio.2010.166

Wallingford, J. B., Niswander, L. A., Shaw, G. M., & Finnell, R. H. (2013). The continuing challenge of understanding, preventing, and treating neural tube defects. *Science*, *339*(6123), 1222002. doi: 10.1126/science.1222002

Williams, I. A., Shaw, R., Kleinman, C. S., Gersony, W. M., Prakash, A., Levasseur, S. M., & Glickstein, J. S. (2008). Parental understanding of neonatal congenital heart disease. *Pediatric Cardiology*, *29*(6), 1059-1065. doi: 10.1007/s00246-008-9254-8

World Health Organization. (2010). Birth defects: report by the secretariat. *Sixty-(2) Third World Health Assembly A*, *63*. Retrieved from http://apps.who.int/gb/ebwha/pdf_files/WHA63/A63_10-en.pdf

Table 1. Information about the articles included in the Literature review

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Hunfeld *et al.*	1999	The Netherlands	Congenital disorders of the digestive tract and central nervous system.	Cross-sectional. Quantitative methods (self-report questionnaire)	16 (50%)	36 (47%)	Impact on Family Scale (IFS) (parental burden) Perinatal Grief Scale (PGS) (Grief over the loss of a healthy child) Functional Health Status Scale (FSII-R) (perception of the mother of the health of her child)	-No difference between mothers and fathers for burden or grief. - Mothers reported more personal strain - One year after infant's birth, mothers in fetal group (FM) showed greater burden, social impact, grief, difficulty with coping, and despair compared to mothers in postnatal group (PM). FM reported lower perceived performance of their child due to their condition - No differences in psychological responses between fathers in fetal (FF) or postnatal (PF) groups ; however, FF showed a trend of greater financial burden - Greater burden and grief associated with fetal diagnosis, low perceived functional health status, and multiple congenital anomalies.	- Small sample size - Heterogeneity in sample
Davalbhakta & Hall	2000	UK	Cleft lip diagnosis	Cross-sectional. Mixed methods (Quantitative and Qualitative) - (self-report questionnaire)	25	65	Questionnaire	- Eighty five per cent of parents who received a fetal diagnosis indicated this helped them psychologically prepare for the birth of their baby - Ninety two per cent of parents said they had not considered pregnancy termination - Irrespective of time of diagnosis, parents perceived the counselling they received as clear and detailed.	- Greater proportion of th sample diagnosed postnatally. Particularly parents of children with isolated cleft of the palate with only 8% diagnosed during pregnancy - Questionnaire utilised was not validated

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Hoehn et al.	2004	USA	Congenital heart disease	Longitudinal,mixed methods design including focus groups, self- report measures (phase 2) & semi-structured interview (phase 3)	Phase 2 - 29 (45%) Phase 3 – 24 (42%)	Phase 2 -23 (39%) Phase 3 – 20 (35%)	- Focus groups (to direct issues discussed in interviews) - Spielberger State/Trait Anxiety Inventory (anxiety) - Life Orientation Test (dispositional optimism) - Life Experiences survey (perceptions of life experiences) - semi-structured interviews	*Phase 2*- No significant difference found in mothers due to timing of diagnosis. FF had less anxiety, more optimism and reported fewer negative life events than PF. *Phase 3*- all groups felt they made a genuine, informed choice for baby to have surgery. No parent expressed regret about decision for surgery - FF were more inclined to express concerns about family finances	- Strong heterogeneity in the sample (lots of differe diagnoses which may account for differences) - Selection bias e.g., only parents of neonates prenatally diagnosed who CONTINUED with pregnancy were included
Skari et al	2006	Norway	Congenital malformations	Longitudinal. Quantitative methods (self-report questionnaire completed 0-7 days after admission for surgery, 6 weeks after birth and 6 months after birth).	107	186 *Also 249 parents of healthy babies were included as a comparison group.*	General Health Questionnaire (Psychological distress) State Anxiety Inventory (anxiety) Impact of Event Scale (Stress-related cognition and behaviour)	(0-7 days)- Fetal parents reported higher psych distress, anxiety and depression than parents with postnatal diagnosis - Mothers scored significantly higher then fathers on all psychological response measures. (6 weeks) – Fetal parents reported increased levels of anxiety, depression and psychological distress (6 months)- No differences between fetal and postnatal parents. Mothers reported higher anxiety than fathers. - Psychological distress was highest if fetal detection was between weeks 25 and 30	- Includes multiple different congenital malformations with varying severity - Selective bias. Those wl didn't respond may have more severe stress response. - Did not include parents of children who died befo admission to the referral centre
Brosig, et al.	2007	US	Congenital heart disease	Longitudinal. Mixed methods. (self-report questionnaires completed at three time-points (diagnosis, birth, 6 months postpartum) and semi-structured interviews at time of diagnosis and 6 months after birth.)	20 (50%)	14 (50%)	- Brief Symptom Inventory (global symptoms of distress)	- Seventy one per cent of postnatal parents and 75% of fetal parents had BSI scores in the clinically significant range at time of diagnosis - At diagnosis, no difference in BSI scores between fetal and postnatal groups. - Six months after birth, fetal group had higher BSI scores - No clinically significant differences were found between mothers and fathers at any time point - Depression scores in prenatal group did not change from diagnosis, to birth to 6mths after - Utilising a chi-squared analysis it was found that parents with child with severe disease were significantly more likely to have BSI scores in clinical range - Emerging themes at both time points were similar in each group.	- Small sample size - Infants in fetal group ha more severe disease compared to postnatal. - Parental adjustment is affected by multiple facto and cannot be attributed t just severity of CHD or timing of diagnosis - Distinct difference in response rate with 91% ir fetal and 44% in postnata group.

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Nusbaum et al	2008	US	Cleft diagnosis	Cross-sectional. Qualitative design (semi-structured interview conducted by a genetic counselling Master's degree student)	12 (33%)	8 (38%)	- Demographic info - Open-ended interviews – emphasis on thematic analysis. Only one set question – 'Tell me the story of how you first found out about your child's cleft". Then questions were asked based on the clients lead.	- Five themes expressed by both fetal and postnatal groups: (1) receiving the diagnosis (varying opinions) 2. shock 3. cause and embodied knowledge 4. Parent to parent support 5. Disability - In addition, 4 themes prewire unique to the fetal group: 1. Coping 2. Preparation 3. Disadvantages of fetal diagnosis 4. Alternative perspectives. - All participants in fetal group reported being 'satisfied' with receiving their baby's diagnosis antenatally.	- The sample lacked diversity e.g., gender - The sample only involve children with an isolated cleft diagnosis excluding children where the cleft may be a consequence of genetic syndrome/chromosome abnormality. - Authors suggested some of the questions in interviews may have been leading.
Williams et al	2008	US	CHD	Cross-sectional. Quantitative methods (self-report questionnaire)	26	24	- Parent Understanding Questionnaire (parental understanding of diagnosis of their baby)	- Fetal diagnosis associated with increased parental understanding at NICU discharge (independent of maternal education) - Association between fetal diagnosis and increased concern about taking the baby home; parents in fetal group 4 times more likely to report worry) - Even with fetal diagnosis, gaps in parental understanding were identified.	- Small sample size - Ascertainment bias as only parents who were physically in the NICU a couple of days before discharge - Measures weren't validated
Skreden et al	2010	Norway	'Malformation'	Longitudinal. Quantitative methods (self-report questionnaire). Measures taken at Phase 1 (0-7 days), phase 2 (6 weeks), phase 3 (6 months) and phase 4 (9 years)	84	134	General Health Questionnaire (anxiety/insomnia, depression, somatization and social dysfunction. State Anxiety Inventory (anxiety), Impact of Event Scale (cognitive/behavioural aspects of stress)	Phase 4- no significant differences in parental psychological responses at between parents due to timing of diagnosis - Mothers rated their child's daily functioning similarly to the fathers - Clinically important psychological distress was reported by 30.2% of parents at long-term follow-up whereas 27.8% reported clinically important state anxiety. - Mothers reported significantly higher scores for overall psychological distress and intrusion than fathers.	- Did not include parents children who died before admission to referral centr - Language limitations – Must speak Norwegian - Included multiple different diagnoses - Hard to transfer results across to poorer countries with lower social benefits and poorer living.

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Robbins et al,	2010	US	Orofacial Clefts	Cross-sectional. Qualitative methods (Structured interview)	46	189	Structured interview conducted with mothers of children ranging from 2 to 7 years of age	- Most mothers expressed satisfaction with information and support received after diagnosis is made - FM were more likely to rate medical staff as moderately or very helpful in making it easier for the infant to feed - No difference in treatment provided by multidisciplinary team (level of treatment didn't vary) - FM of children diagnosed with cleft lip and palate were significantly less satisfied with their child's appearance at 2 to 7 years of age - FM somewhat more likely to report their child was having difficulty being understood at 2 to 7 years of age	Greater proportion of the sample diagnosed postnatally with only 19.6% of mothers receivi a fetal diagnosis.
Berggren et al	2012	Sweden	Cleft diagnosis	Cross-sectional. Quantitative methods (self-report questionnaires)	36	46	Questionnaires not validated (demographics and opinions regarding counselling offered)	- Explored parents experiences fetal counselling for cleft palate. - Fifty six per cent of parents in postnatal group would have wanted to have received a fetal diagnosis	- Information was cross-sectional, longitudinal study would have been more informative - Measures are not validated - Study is retrospective - Other possible factors n considered e.g., crisis reaction, severity of malformation etc
Fonseca, Nazare and Canavarro	2012	Portugal	Congenital anomaly including: unrinary system anomalies, CHD, visible anomalies, nervous system anomalies, digestive system anomalies.	Cross-sectional. Quantitative methods (self-report questionnaire)	42 couples in the clinical group (50%)	42 couples in the comparison group (50%)	Brief Symptom Inventory (psychological distress) WHOQOL (Quality of life) Demographics	- No differences between fetal and post groups in psychological distress - Fetal diagnosis associated with higher maternal psychological QoL - Parents of a baby with a congenital anomaly reported greater psychological distress compared to parents of healthy babies. - Women presented with significantly higher levels of anxiety and depression. - Mothers presented with significantly lower QoL than fathers on psychological and physical domains.	- Small sample size therefore small effects could not be detected - Multiple congenital anomalies included/not differentiated

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Bevilacqua et al	2013	Italy	CHD	Cross-sectional. Quantitative methods (self-report questionnaire)	40 (50%)	36 (50%)	General Health Qu (GHQ-30) (30 items) (General health, sleep, psychological distress) Beck Depression Inventory (21items) (Depressions Health Survey-36 (36 items) (Health related QoL)	- No difference in stress and depression were found due to time of diagnosis - Stress and depression levels significantly higher in mothers (81.8% stress and 45.6% depression) than fathers (60.6% stress and 20.0% depression) - In percentage, FM were more depressed and PM were more stressed	- Small sample size - Lacks statistical power - Measurements limited due to self-report measure
Fonseca, Nazare and Canavarro	2013	Portugal	Congenital anomaly including: CHD, nervous system anomalies, digestive system anomalies, urinary system anomalies, oro-facial clefts, limb abnormalies.	Cross-sectional. Quantitative methods (self-report questionnaire)	60 mothers involved (54.5%)	50 fathers involved (45.5%)	Demographics 'When you learned of your infant's diagnosis, how much did you feel the following emotions?' (scale 0-100)	- Results indicated that mothers and fathers had comparative emotional responses. This means that mothers and fathers were comparative within each cluster. - In 72% of families, both parents had similar patterns of emotional response to dx - Type of congenital anomaly affected the intensity of emotion in mothers but not fathers. In mothers, the type of congenital anomaly affected the intensity of sadness, guilt and anger - When looking at the intensity of the emotion, mothers in fetal group felt significantly more anger and sadness than mothers in postnatal - Fathers who had no prior knowledge about the congenital anomaly showed a pattern of high intensity negative emotions (therefore needed to learn what the anomaly was by gathering information about the diagnosis) - Greater uncertainty about the diagnosis was associated with more intense emotional responses at disclosure.	- Small sample size - Retrospective assessmen of emotional reaction - Measures utilised are no validated

Authors	Year	Country	Disease group	Study Design	Fetal dx *n* (% fathers)	Postnatal dx *n* (% fathers)	Measures	Key findings	Key Limitations
Nelson Goff et al	2013	US	Down Syndrome	Cross-sectional. Qualitative methods (web-based survey)	46 (6.5%)	115 (10.4%)	- Four prompting questions on online survey	- Three main themes identified: (1) fetal screening/testing decisions by parents, (2) adjustment for parents, and (3) post diagnosis resources and support for parents. - Participants in both groups identified having negative experiences with medical professionals during diagnosis	- Very specific sample utilised e.g., married, white, high socioeconomi status - Severity of impairment c multiple diagnoses were not reported/addressed in the sample - Small representation of fathers
Fonseca et al	2014	Portugal	Congenital anomaly including: CHD, nervous system anomalies, digestive system anomalies, urinary system anomalies, oro-facial clefts, limb anomalies.	Longitudinal. Quantitative methods (self-report questionnaire completed one month after diagnosis (time point 1) and six months after birth (time point 2)	Proportion of fetal and postnatal not specified. Instead reported- 43 mothers (54.5%), 36 fathers (45.5%)		- Brief Symptom Inventory (Psychological distress) - WHOQOL-Brief (QoL) - socio-demographic and clinical information	- Study found no significant effects of timing of diagnosis at either time points. – No gender differences were found in the fetal diagnosis group at either time point - Time point 1- mothers were found to have higher anxiety and depression than fathers. - Time point 2 - mothers were found to have higher anxiety but not depression than fathers. - Also found effects for QoL with mothers having lower physical and psychological QoL than fathers at time point 1 and lower psychological QoL at time point 2. - Postnatal mothers found to have higher social relationships QoL than their partners - All but one parent showed a pattern of recovery from T1 to T2.	- Small sample size - Heterogeneity in the sample

Table 2. Psychological constructs measured and psychometric tools used in the 15 studies identified in this review.

Psychological Constructs measured	Psychometric tools used	Frequency of use
Psychological distress	General Health Questionnaire (GHQ-28)	Skari et al., 2006; Skreden et al., 2010; Bevilacqua et al., 2013.
	Brief Symptom Inventory	Brosig et al., 2007; Fonseca et al., 2012; Fonseca et al., 2014.
Anxiety	State Anxiety Inventory (STAI-XI)	Skari et al., 2006; Skreden et al., 2010
	Spielberger State/Trait Anxiety Inventory	Hoehn et al., 2004
Quality of life	World Health Organisation Quality of Life-Brief (WHOQOL-Bref)	Fonseca et al., 2012; Fonseca et al., 2014.
	Health Survey-36	Bevilacqua et al., 2013.
Family burden	Impact on Family Scale (IFS)	Hunfeld et al., 1999
Grief	Perinatal Grief Scale (PGS)	Hunfeld et al., 1999
Parent's perception of their child's health	Functional Health Status Scale (FSII-R)	Hunfeld et al., 1999
Traumatic stress	Impact of Events Scale (IES)	Skari et al., 2006; Skreden et al., 2010
Depression	Beck Depression Inventory	Bevilacqua et al., 2013.
Optimism	Life Orientation Test	Hoehn et al., 2004
Perception of experiences in life	Life experiences survey	Hoehn et al., 2004
Understanding of diagnosis	Non-validated measure	Williams et al., 2008
Experiences of counselling	Non-validated measure	Berggren et al., 2012
Emotional reaction to diagnosis	Visual analogue scale from 0 ('I did not feel it at all') to 100 ('I felt it a lot')	Fonseca et al., 2013

Manuscript: Parental Coping After Their Baby's Diagnosis of Congenital Heart Disease

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Abstract

In our study we investigated the experiences of mothers and fathers who had received either a fetal or postnatal diagnosis of complex congenital heart disease (CHD) in their baby, specifically exploring their methods of coping. Twenty-five mothers and fathers (six fetal mothers, six fetal fathers, nine postnatal mothers, four postnatal fathers) participated in semi-structured interviews which we analyzed utilizing a modified analytic induction approach. In our analysis we explored the applicability of the model of stress and coping proposed by Lazarus and Folkman (1984) to these parents and their methods of coping and found the majority of themes communicated by parents could be classified within the model, with the exception of parental pride and focus on the baby. We also explored differences due to time of diagnosis, finding that fetal and postnatal diagnostic groups differed in level of preparedness, distancing, meaning-focused coping and sources of reassurance. Differences between mothers and fathers were also investigated, with differences in appraisal, confrontive coping, use of social support and meaning-focused coping identified.

Key words: caregivers/caregiving, childbirth, children, illness and disease coping and adaptation, families, fathers, heart health, interviews, semistructured, mothers, pediatrics, pregnancy, qualitative analysis

Becoming a parent is a significant life event, impacting upon all facets of life, changing an individual's point of view and at times, leaving parents in a state of shock and dislocation soon after their baby's birth (Pancer, Pratt, Hunsberger & Gallant, 2000; Tracey, 2000). Consider then the additional psychological impact on parents who receive a diagnosis of complex congenital heart disease (CHD) for their baby. This is a reality many parents must face, with reports indicating that CHD affects over 2,000 babies born each year in Australia (Blue, Kirk, Sholler, Harvey & Winlaw, 2012). The term "congenital heart disease" encompasses "any disorder of the heart or central blood vessels that is present at birth and can range in severity from minor murmurs to severe abnormalities" (Australian Institute of Health and Welfare, 2011, p. 127). For the purposes of this research, a diagnosis of complex CHD is indicative of any structural congenital heart abnormality requiring surgery during the first six months of life.

Receiving a diagnosis of complex CHD for one's baby is a highly distressing and potentially traumatic experience, and can have resounding psychological implications (Menahem & Grimwade, 2005; Howland, 2007; Rychik et al., 2013). The literature indicates that parents of children with complex CHD experience elevated levels of stress, anxiety, and despair (Soulvie, Desai, White & Sullivan, 2012). For example, when compared to parents of children with other diseases or to parents of healthy children, parents of children with CHD demonstrate significantly greater distress and hopelessness (Lawoko & Soares, 2002). In a sample of fifty-nine mothers, assessed two-four weeks after receiving a fetal diagnosis of CHD, 39% reported levels of traumatic distress warranting clinical intervention, 31% reported clinically-significant state anxiety, and 22% reported depressive symptoms indicative of a need for clinical intervention (Rychik et al., 2013). Parents have reported similar

levels of distress after receiving a postnatal diagnosis of CHD (Doherty et al., 2009). In a sample of 140 participants, completing assessments an average 2.8 months after the birth of their newborn, one-third of mothers and 18% of fathers fell within the clinical range for psychological stress, including anxiety and depression (Doherty et al., 2009).

As indicated above, it is currently possible to receive a diagnosis of complex CHD at two different time points; during pregnancy (fetal diagnosis) or after the birth of the baby (postnatal diagnosis). As technology continues to advance, the likelihood of fetal diagnosis has increased (Sholler, Kasparian, Pye, Cole & Winlaw, 2011). For example, at the Heart Centre for Children at the Children's Hospital at Westmead, approximately 44% of cases of complex CHD addressed are detected during pregnancy (Sholler, Kasparian, Pye, Cole & Winlaw, 2011). There is still debate, however, about the psychological impact of the timing of cardiac diagnosis. A recent review of the literature exploring studies that considered all types of congenital anomalies (see Part 1 of this thesis) found conflicting results, with 40% of studies finding no difference in psychological distress amongst parents as a result of time of diagnosis (fetal versus postnatal). In contrast, multiple studies have reported better psychological outcomes for parents who receive a fetal diagnosis (Fonseca, Nazare & Canavarro, 2012; Hoehn et al., 2004), whilst other studies indicate better outcomes for parents who receive a postnatal diagnosis (Skari et al., 2006). The factors underlying these mixed findings are unclear. It has been suggested that a fetal diagnosis is advantageous as it provides parents with the time and opportunity to consider treatment options for their baby and make a decision regarding the possibility of pregnancy termination (Rychik et al., 2013). However, it has also been speculated that a fetal diagnosis potentially acts as an ever-present psychological

stressor for parents, particularly when the condition identified cannot be treated until after birth (Skari et al., 2006). This raises the possibility that a postnatal diagnosis could lead to better psychological outcomes for parents, as they learn of the diagnosis at a time when something can be actively done about it (Skari et al., 2006).

Also contentious is whether mothers and fathers respond differently to complex CHD diagnosis. Only relatively recently have fathers' experiences been considered and measured in research in the context of paediatric illness and thus, it is difficult to determine if such differences exist (Doherty et al., 2009; Brosig et al, 2007). In a recent review of the literature, over 75% of published studies found mothers report greater psychological distress after diagnosis of congenital anomaly in their baby than fathers (see Part 1 of this thesis). Due to limited research exploring this area, reasons for this difference in distress is currently conjecture. One possible explanation is in reporting, with fathers less inclined than mothers to accurately disclose the nature of their experience and their emotions (Mirowsky & Ross, 1995). These findings may arise due to gender differences in the appraisal of the severity of the stressful situation, with women, when presented with the same situation as men, found to report the situation as more stressful than their male counterparts (Eaton & Bradley, 2008). Also of consideration are cultural gender roles and expectations and the possibility that mothers simply did experience greater distress than fathers.

When faced with a diagnosis of complex CHD in their baby, mothers and fathers, regardless of time of diagnosis, need to find a way in which to cope. In our article we will investigate the coping strategies employed by parents as they try to cope with the diagnosed condition and required treatment.

History and Definition of Coping

Coping is a complex concept, difficult to define and quantify (Beutler, Moos & Lane, 2003). It was first introduced into the psychological literature though Freud's psychoanalytic work on defence mechanisms in the 1920's and 1930's (Livneh and Martz, 2007). While it is not within the scope of this review to extensively outline these defence mechanisms, his work, further developed by Anna Freud, inspired the initial conceptualisations of person based coping (Livneh & Martz, 2007). Distinctions between coping and defence mechanisms were drawn by later researchers including Norma Haan and Theodore C. Kroeber, who devised a model of ego functioning and claimed that coping was a parallel process to defence processes but was more "flexible, purposive, reality oriented and differentiated" (Haan, 1965, p 374).

Coping became a more prevalent topic in the psychological literature in the 1960's and 1970's, studied in conjunction with the popular topic of stress (Zaumseil & Schwarz, 2014). Since this time, a plethora of models of stress and coping have emerged and have attempted to address and take into account the interaction between the individual and the environment in which they are trying to cope (Livneh and Martz, 2007). Two prominent theoretical approaches to understanding and conceptualising coping include Hobfall's model of Conservation of Resources (1989) and Lazarus and Folkman's model of Stress and Coping (1984). The central premise of Hobfall's model of Conservation of Resources (1989) model is that an individual aim to attain, defend and retain resources, and stress is anything that threatens to deplete or damage those resources (Hobfall, 1989). Within this model, coping involves employing resources to combat threats, which in turn leads to depletion of valuable resources (Hobfall, 1989). It is therefore possible to determine if the method

of coping implemented is beneficial or problematic by weighing up the resources protected and the resources utilised while trying to combat threats (Hobfall, 1989). One fundamental element of the model is the assumption that one individual will perceive a loss of resources comparably to another person when faced with a similar situation (Hobfall, 2001). The model also places great significance in culture, stipulating that culture determines the level of importance given to each of the different resources (Hobfall, 2001). Consequently, the model does not adequately take into account differences between individuals.

In 1984, Lazarus and Folkman posited a model of stress and coping that is still widely recognised and utilised in clinical and health psychology research and that recognises individual differences in the experience of stress and coping (Folkman, 2010). Lazarus and Folkman's model of stress and coping is transactional, acknowledging a bidirectional interaction between the individual and his or her environment (1984). One of the key strengths of the model is the recognition of individuality and the perspective that people bring their own unique resources and coping strategies to functioning within a dynamic and stressful situation (Frydenberg, 2014). Lazarus and Folkman propose that stress is a process that can be modified by the individual and variation in coping can be observed over time (Quine & Pahl, 1991). The model dictates that two primary processes mediate this relationship between the individual and his or her environment: cognitive appraisal and coping (Folkman, 2010).

Cognitive appraisal involves the individual formulating an evaluation of the situation, first conducting a primary appraisal to determine if the situation is significant and poses a threat to the self or to their loved ones (Folkman, 2010). This process is influenced by individual attitudes, values, priorities and ambitions

(Folkman, 2010). This process is then followed by a secondary appraisal (Folkman, Lazarus, Dunkel-Schetter, DeLongis & Gruen, 1986), involving an evaluation of the options available to the individual to change, prevent, control or cope with the situation (Folkman, 2010).

Once this appraisal process is complete, the individual has achieved a final assessment of the implications of the situation. Of foremost concern is whether the situation poses a potential threat or whether it provides a challenge and opportunity for growth (Folkman, 2010). The result of the appraisal evokes varying emotions. For example, a threat causes fear and concern whilst a challenge arouses excitement and anticipation (Folkman, 2010).

According to the model, the secondary process mediating the bidirectional relationship between the individual and his or her environment when combating stress is coping (Folkman, 2010). Coping is defined as "the thoughts, [feelings] and behaviours people use to manage the internal and external demands of stressful events" (Folkman, 2010, p 902). In the initial model, two broad types of coping were identified and outlined; problem-focused coping and emotion-focused coping (Lazarus & Folkman, 1984). Problem-focused coping involves taking active, analytic and task-oriented steps to solve or manage a stressful situation through gathering information, obtaining required resources, formulating decisions and plans, and problem solving (Folkman, 2010). Emotion-focused coping involves regulating the emotions evoked by a distressing situation through various strategies such as distancing, escape-avoidance, or seeking social and emotional support (Folkman, 2010).

More recently, Folkman and Moskowitz have proposed the inclusion of a third broad type of coping in order to acknowledge the role of positive, as well as

negative, emotions in the stress and coping process (2000). Folkman defined meaning-focused coping as a way that an individual views a difficult situation, drawing upon their own beliefs, values, goals and attitudes to find purpose in that situation and as a consequence be able to cope with the difficult situation (Folkman, 2008). She theorised that meaning-focused coping helps to restore coping resources and consequently ensures continued problem-focused coping by inciting positive emotions and positive appraisals of the situation (Folkman, 2008). This type of coping is particularly pertinent in situations that cannot be readily resolved such as illness, assisting the individual to avoid falling into a cycle of chronic stress (Folkman, 2008). Five categories of meaning-focused coping are described within the model, including benefit finding, benefit reminding, adaptive goal processes, reordering priorities, and infusing ordinary events with positive meaning (please refer to Appendix A for a definition of each type of coping category) (Folkman, 2008).

In terms of potential gender differences in coping, previous research has hypothesised that men tend to employ problem-focused coping when faced with a stressful situation, while women are more inclined to utilise emotion-focused coping, particularly expression of emotions and seeking social support (Melendez, Mayordomo, Sancho & Tomas, 2012). Recent studies have, however, produced somewhat contradictory findings, with women observed to score more highly on emotion-focused coping strategies than men, whilst also reporting a comparable use of problem-focused coping strategies (Maltaud, 2004; Melendez et al., 2012). Further, in a review of the literature, it was found women were more likely to engage in most coping strategies, particularly those that involved communicating with others or to self such as seeking emotional support, than men, and were also found to

engage more frequently in meaning-focused coping such as positive reappraisal (Tamres, Janicki and Helgeson, 2002).

Parental coping after their baby's diagnosis of congenital heart disease

A key objective of our article is to apply Lazarus and Folkman's model of stress and coping to the experiences of mothers and fathers who have received a diagnosis of complex CHD in their baby in order to highlight the coping behaviours frequently employed within this particular population (1984). Currently, extensive research exists documenting the psychological impact of such a diagnosis yet only a limited number of studies have explicitly explored the different coping strategies utilised by parents in their attempts to cope with their baby's diagnosis of heart disease.

Emotion focused coping

In a review of the literature, it is evident that receiving a diagnosis of CHD in your infant elicits a wide range of different and, at times, conflicting emotions. In a qualitative study of parents of an infant with hypoplastic left heart syndrome, mothers and fathers reported having difficulty balancing their fears for their child's health with their pride in their child's strength and resilience (Lee & Rempel, 2011). Similarly, Clark and Miles (1999) found that fathers experienced significantly conflicting responses after becoming a father to an infant with CHD. They reported excitement in becoming a father but sadness due to the infant's condition, expressed difficulties in building a bond with the infant but also recognising the infant's frailty and trying to be strong emotionally and take control while experiencing strong emotions and feeling helpless (Clark & Miles, 1999). Recently, Harvey, Kovalesky, Woods and Loan (2013) identified six main themes in the experiences of eight mothers of children with CHD at three different time points: before, during and after

surgery. Mothers completed journal entries and their children ranged in age from 7 days old at first surgery to 9 years old at time of last surgery (Harvey et al., 2013). All mothers reported experiencing a wide range of intense emotions and all spoke about the challenge they faced in trying to maintain their role as mother amongst the chaos of treatment (Harvey et al., 2013). Mothers reported feeling uncertainty surrounding their child's future and expressed real difficulty in accepting the chance that their baby may not survive (Harvey et al., 2013). Only in Lee and Rempel's 2011 study were coping strategies identified with normalisation identified as the primary strategy employed by parents as they tried to reconcile such conflicting attitudes and emotions. This finding suggests that as parents try to balance their pride, their fears and their various emotional reactions to their babie's diagnosis they focus on recognising and treating their child as a normal child, fighting against allowing them to be defined by their heart condition and as a consequence building resilience in their role as parents (Lee & Rempel, 2011).

Emotion focused coping – Social support

Social support has been identified as an important resource for parents when trying to cope (Tak & McCubbin, 2002; Svavarsdottir & McCubbin, 1996; Spijkerboer et al., 2007; Doherty et al., 2009). For example, Tak and McCubbin (2002) identified perceived social support as a significant indicator of coping in parents of children with heart disease aged less than 12 years.

Further, when CHD mothers and fathers were compared to a reference group of the Utrecht Coping List measure, composed of railway employees, nurses and members of the general population, coping strategies employed were largely comparative (Spijkerboer et al., 2007). It was found that CHD mothers were more inclined to report seeking out social support and both CHD mothers and fathers were

less likely to employ expression of negative emotions and reassuring thoughts as means of coping (Spijkerboer et al., 2007). Finding little difference between the reference group and parents of children with CHD may be indicative of general similarities across populations and circumstances. If so, it may be difficult to identify differences amongst parents who receive either a fetal or postnatal diagnosis.

Emotion focused coping – Family support

Family support has also been highlighted in the literature as an important coping strategy employed by parents of infants with a diagnosis of CHD (Sira, Desai, Sullivan & Hannon, 2014; Svavarsdottir & McCubbin, 1996; Doherty et al., 2009). In a recent study, mothers who had a tendency to report coping strategies emphasising family integration, drawing on family for support and taking an optimistic outlook were found to also to report high levels of spirituality (Sira et al., 2014). Sira et al. (2014) speculated that one of the ways in which mothers achieved family integration was through spirituality. Interestingly, the research found mothers were much less likely to maintain social relationships outside of the family unit and less likely to engage in self-care behaviours (Sira et al., 2014). In a more recent study, Doherty et al. (2009) explored psychological functioning amongst mothers and fathers after receiving a diagnosis of major CHD in their newborn, and found that mothers reported significantly higher levels of psychological distress than fathers (33% of mothers in the clinical range on the Brief Symptom Index compared to 18% of fathers). The study also compared the coping strategies employed by mothers and fathers and found that mothers used instrumental social support, emotional social support and religion more than fathers, whilst fathers reported using alcohol significantly more frequently than mothers (Doherty et al., 2009). Consequently, the

utilisation of family support as a coping strategy has been identified in mothers but has yet to be conclusively reported by fathers.

Problem-focused coping – Information gathering

Contrary to previous hypotheses in the literature, several studies identified problem-focused coping as an important coping strategy for mothers of children with CHD (Sira et al., 2014; Svavarsdottir & McCubbin, 1996; Davis, Brown, Bakeman & Campbell, 1998). For instance, in a recent survey of 175 mothers of children with CHD, Sira et al. (2014) found mothers reported they needed to understand the medical condition of their child and as a consequence, an important coping strategy was to use the Internet to gain further information and to connect with other parents of CHD children (Sira et al., 2014). Similarly, Davis, Brown, Bakeman and Campbell (1998) reported that maternal adjustment of mothers of children with CHD was correlated with active strategies for coping. Interestingly, Tak and McCubbin (2002) found that parental age was a relevant predictive factor in the use of problem-focused coping with younger mothers and fathers utilising more helpful coping such as greater use of social support and access to medical information than their older counterparts.

Moreover, in a study that compared the coping strategies of mothers and fathers, Svavarsdottir and McCubbin (1996) found mothers more frequently reported gathering medical information, forming relationships with medical staff and other parents of a child with CHD, and actively seeking to understand the medical condition of their child compared to fathers. This finding may be indicative of the employment of problem-focused coping by mothers but it may also be a reflection of the different roles played by mothers and fathers and the different opportunities they are afforded after receiving a diagnosis in their baby. Mothers more frequent use of

problem solving coping and formation of relationships with medical staff may be due to practical causes such as fathers needing to leave the hospital due to work, looking after other children in the family or running errands while mothers stay by their baby's bedside. In this situation it would be difficult for fathers to employ the same level of problem-focused coping specifically surrounding the babies diagnosis due to a lack of access to the medical team. These findings and considerations highlight the need to examine the use of the coping strategies within a wider context, particularly when comparing different groups such as mothers with fathers.

Meaning-focused coping

Meaning focused coping has also previously been identified in recent research (Lee & Rempel, 2011; Harvey et al., 2013). Lee & Rempel (2011) found parents of infants with hypoplastic left heart syndrome frequently employed a positive outlook and drew positive meaning from their experiences. It is important to further recognise that while parents were able to draw this meaning, they also expressed continued fears about their child's health and frustrations in their inability to always protect their child (Lee & Rempel, 2011). In their qualitative study Harvey et al. (2013) found that all eight mothers who participated were able to find meaning in their experience, with several reporting their spirituality had been strengthened (Harvey et al., 2013). These findings indicate that while parents are facing a very emotional and challenging experience in receiving a diagnosis of CHD in their babies they are able to draw meaning and strength from the experience (Lee & Rempel, 2011; Harvey et al., 2013).

As evidenced above, the current literature is sparse in its exploration of coping strategies employed by mothers and fathers in the acute and short-term period after receiving a diagnosis of CHD in their baby. Further, of studies that have investigated

this population, results are currently varied and conflicting. Limited studies have specifically investigated differences in coping as a result of time of diagnosis and gender and we were unable to find any other research that had explored the applicability of Lazarus and Folkman's model of stress and coping (1984), including also Folkman and Moskowitz's recent additions of meaning-focused coping (2000). Thus, aims of the current article were threefold. First, to explore whether, after receiving a diagnosis of CHD in their baby, parents respond to such a diagnosis in a manner consistent with the stress and coping model proposed by Lazarus and Folkman (1984) with additional revisions made by Folkman and Moskowitz (2000). Second, whether there are clear thematic differences in the ways in which parents describe their coping responses after receiving either a fetal or postnatal a diagnosis of complex congenital heart disease in their baby. Finally, to investigate whether the patterns of coping described are different for mothers when compared to fathers.

Methodology

Study Design

A cross-sectional, mixed methods study design was utilised, involving the collection of both qualitative and quantitative data. This design allowed for a wide-ranging exploration of a topic about which there is a dearth of research and provided a better chance to attempt to understand the complexities of the experience of parents after receiving their baby's diagnosis of complex CHD (Bazeley, 2013). In addition, various disciplines were represented on the research team, including psychology, fetal and paediatric cardiology, paediatric cardiothoracic surgery, medical genetics and infant and perinatal psychiatry, as well as two parent representatives (one mother, one father).

Participants

Study participants included mothers and fathers of a baby diagnosed with complex congenital heart disease (CHD) either during pregnancy (fetal diagnosis) or within the first six months of life (postnatal diagnosis) (Figure 1). All diagnoses were received between September 2011 and September 2012, and for the purposes of this study, complex CHD was defined as any structural congenital heart abnormality requiring surgery during the first six months of life. Participants were identified through the cardiology databases of the Sydney Children's Hospitals Network, including sites at both the Children's Hospital at Westmead and the Sydney Children's Hospital, Randwick. In order to gather information from as many different perspectives as possible and generate a diverse sample, maximum variation sampling was utilised, including babies with a variety of cardiac diagnoses, and parents from a range of residential locations, and with varying beliefs and experiences (Miles & Huberman, 1994). Further, the sample included a comparative number of parents who received a fetal or postnatal diagnosis. Parents were also asked to participate at varying periods of their baby's medical journey, ranging from pregnancy through to their infant's first birthday.

To be eligible to take part in the study, parents were required to provide informed consent, be over the age of 18, and be able to complete the study interview and questionnaire in English. Parents were not excluded on the basis of marital status.

Procedure

Ethics approval was received from all relevant Human Research Ethics Committees (HRECs), including the University of Wollongong (Approval: HE08/132), the Sydney Children's Hospital Randwick, the Children's Hospital

Westmead and the University of Newcastle (Approval: H-2015-0012). A free-call telephone line was set up at the beginning of the study for participants to utilise for any queries or concerns.

Identified eligible participants were initially contacted via mail and were sent a study package (See Appendix B). This included an invitation letter from their baby's treating paediatric cardiologist, a participant information sheet, consent form, and a reply paid envelope. Reminder letters and telephone calls were made to participants who did not respond to the mail out within two weeks. In accordance with ethics guidelines, attempts to contact families were no longer made after one telephone conversation and the study package was sent a second time.

Informed written consent was collected from all parents and, once consented, participants were asked to indicate their preferences in terms of interview time, venue (home, hospital, over the telephone), and format (individual or with their partner). Any study-related travel expenses incurred by parents were reimbursed. One week after the interview had taken place, participants were sent a paper-based self-report questionnaire to complete and mail back to the research team.

Qualitative Data Collection and Analysis

Semi-structured interviews were undertaken and analysed using the framework outlined by Miles and Huberman (2002). The principal investigator, Dr Nadine Kasparian, conducted all interviews and the interview process was guided by pre-formulated questions outlined in two separate Discussion Guides (Fetal and Postnatal) created by the research team (See Appendix C). The role of the interviewer was to facilitate the discussion, asking questions to encourage parents to openly discuss their feelings, thoughts, experiences and hopes for the future after

receiving their baby's diagnosis. All interviews were audio-recorded with participants' permission and transcribed verbatim.

Transcripts were coded utilising a modified analytic induction approach (Gilgun, 1995). This approach involved formulating initial hypotheses prior to data analysis and then revising these hypotheses to better reflect these data as the analysis took place (Gilgun, 1995). Throughout this process, the researchers were continually looking for evidence to disprove the revised hypotheses, looking for examples of cases that did not conform or confirm the hypotheses (Gilgun, 1995). This approach encourages researchers to seek out variability in the data and assists in the development of broader and more inclusive hypotheses (Gilgun, 1995).

The first step of analysis involved the reading of all transcripts and the generation of brief individual summaries. To achieve high levels of reliability and validity, a multi-level consensus coding method was then employed to code these data. This involved regular meetings of coders to initially identify an appropriate coding system. One coder (HR) then independently coded three transcripts and discussed coding decisions and discrepancies with a second coder (NK). Once generated, this coding system was used in the analysis of the remaining transcripts, with the assistance of the qualitative data analysis software, *QSR NVivo10*. During this coding process, any difficulties in coding or the need for any additional nodes were discussed during weekly coding meetings. After coding was complete, conceptually clustered tables were produced in Microsoft Excel to facilitate the generation of categories and themes found within these data across participant characteristics, including gender (mother or father) and time of diagnosis (fetal or postnatal) (Refer to Table 3). To address potential researcher bias, this table included

counts outlining the number of parents who reflected each particular theme or coping strategy.

Quantitative Data Collection and Analysis

Information collected via self-report questionnaire included demographics (parent age, country of birth, language(s) spoken at home, marital status, education, total gross family income, occupation, number of children, age of children, if they had ever lost an unborn or living child to CHD, and childbearing intentions in the future). Medical information was also collected from cardiology databases, including: infant's age, date of birth, time of diagnosis, age at diagnosis, primary cardiac diagnosis, type of cardiac abnormality, number of cardiac surgeries in the first year of life, age at time of first cardiac surgery, use of pulmonary bypass, whether baby went home before his or her first surgery, and surgical risk according to the Risk Adjustment for Congenital Heart Surgery (RACHS) system. These data have been reported and published elsewhere (Kasparian et al., in preparation).

Results

Demographic characteristics of the sample (N=25)

In the principal study, a total of 53 parents of 31 infants participated with an interview participation rate of 72% and survey participation rate of 89%. Of these, 25 parents of 15 infants took part in the study interview after the birth of their infant and before their infant reached 6 months of age, and these participants comprised the sample for the present sub-study. In this group, 76% of parents took part in the interview individually and 3 couples opted to take part in the interview together, resulting in a total of 22 interviews in this sub-study (Please refer to Table 4 for interview characteristics).

Overall, 60% of participants were mothers and parents who received a fetal (48%) or postnatal (52%) diagnosis were represented almost equally. The fetal diagnostic group consisted of 6 mothers and 6 fathers and the postnatal diagnostic group included 9 mothers and 4 fathers. Over half of the sample (14/25) were married at the time of study participation (Please refer to Table 5 for Demographics of parents).

Infant characteristics

At the time of interview, infants ranged in age from 8 to 184 days (M=103.2 days, SD=63.5). Sixty percent of participants (15/25) were able to take their infants home prior to surgical intervention. The mean time since cardiac diagnosis was 140.6 days (SD=75.6) and significantly differed between the fetal and postnatal diagnostic group ($t(23)=3.694$, $p=0.001$). Mean time since last surgery was 71.2 days (SD=53.2) which did not significantly differ as a result of time of diagnosis ($t(20)=.748$, $p=.463$) (Please refer to Table 6 for further Infant characteristics).

Thematic patterns

Primary appraisal of the diagnosis

After receiving their baby's diagnosis of complex CHD, all parents appraised the situation as personally significant: "I remember I was basically hyperventilating. That was the worst moment of my life, just that realisation." (Please refer to table 7 for main components of Lazarus and Folkman's model of stress and coping identified in data).

Emotions experienced upon receiving the diagnosis ranged from devastation ($n=6/25$), fear ($n=6/25$) and shock ($n=12/25$), through to anger ($n=6/25$), relief ($n=4/25$) and numbness ($n=4/25$). The experience of shock was expressed by a high

proportion of mothers in both diagnostic groups, with only a small number of fathers reporting this emotion (*Mother FDx*=5/6*, Mother PDx*=5/9, *Father FDx*=1/6, *Father PDx*=1/4). "We kind of, you know, sat there just shell-shocked really."

A high proportion of mothers and fathers (*n*=21/25) expressed specific concerns for the baby, including worries about the pain the baby may experience during treatment and fears about the possibility their baby would die (*Mother FDx*=5/6, *Father FDx*=6/6, *Mother PDx*=7/9, *Father PDx*=3/4). "…I remember distinctly thinking that what if he doesn't make it through the surgery? His life might just end. I think that made me very sad."

More than three-quarters of mothers in the postnatal group (*n*=7/9) described the diagnosis as a pivotal event in their lives. This involved descriptions such as "bombshell", "the biggest thing ever" or "the world just fell apart." A smaller proportion of parents in the remaining three groups drew on such descriptions (*Mother FDx*=2/6, *Father FDx* = 1/6, *Father PDx* = 2/4).

Secondary appraisal of the diagnosis

Half of mothers and fathers in the fetal group reported feeling generally prepared for the challenges of their baby's treatment and recovery in contrast with only a small number of parents in the postnatal group expressing a sense of preparedness (*n*=8/25, *Mother FDx*= 4/6, *Father FDx*=3/6, *Mother PDx*=2/9, *Father PDx*=0/4). In contrast, one father who received a fetal diagnosis explained that he did not feel it was possible to be prepared for the birth of the baby with complex CHD:

> You know what? When the baby came I wasn't prepared at all, you know, even though I'd accepted what was going to happen and I knew in my mind what was going to happen, it's still just a step-by-step process.

Several resources were identified as integral in helping parents to feel that were

prepared and equipped to handle the diagnosis. Parents who received a fetal diagnosis more frequently highlighted access to the best medical care available as important in helping them to cope (*n*=8/25, *Mother FDx*=2/6, *Father FDx*=4/6, *Mother PDx*=0/9, *Father PDx*=2/4):

> Like he's a baby that happens to have an abnormal heart and it was sort of that and some other things that sort of certainly helped me get my head around the fact that you know what, this is something that we can, we can deal with, that modern medicine knows how to rectify.

Fathers with a postnatal diagnosis identified having information and knowledge about what was going to happen as important (*n*=6/25, *Mother FDx*=1/6, *Father FDx*=1/6, *Mother PDx*=1/9, *Father PDx*=3/4), and a small number of parents highlighted their supportive partner as an important resource (*n*=4/25, *Mother FDx*=2/6, *Father FDx*=0/6, *Mother PDx*=1/9, *Father PDx*=1/4).

Mothers and fathers from both fetal and postnatal groups conveyed an attitude of trying to "deal with" and accept the diagnosis (*n*=10/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=3/9, *Father PDx*=2/6). For example, one mother explained:

> So for us I think it was like you have to go through it, no matter, you had no choice, this is, this is what you have to do and you have to deal with it, you know, the best you can.

In contrast, a number of parents reported feeling they did not know how they were going to cope. After diagnosis, half of fathers in the fetal group, and almost half of mothers in the postnatal group, described feeling like they did not know what was going to happen or what they were going to do (*n*=8/25, *Mother FDx*=1/6, *Father FDx*=3/6, *Mother PDx*=4/9, *Father PDx*=0/4): "'Oh shit, what are we going to do here? Birth defect and what's it going to do like?'"

Feeling helpless and out of control were themes evident mostly in the narratives of parents who received a postnatal diagnosis (*n*=8/25, *Mother FDx*=0/6, *Father FDx*=2/6, *Mother PDx*=3/9, *Father PDx*=3/4). One mother expressed her frustration in regards to her inability to help her baby and to play the maternal role she had hoped for: "You're kind of, you know, you're supposed to be his mum, you're supposed to be doing things for them, and all of a sudden you're not allowed to do nothing, you can't do nothing."

A small number of parents recalled feeling concerned about their ability to cope with the diagnosis, particularly highlighting concerns regarding access to resources such as money, time and energy (*n*=5/25): "…this baby who we hadn't planned to start off with, and then you know, [we] just spent so much time away from the other kids and expense and everything else, it was just … yeah, a bit hectic."

Methods of Coping

Accepting responsibility

After initially receiving the diagnosis, the majority of parents described searching for an explanation as to why the condition had occurred, questioning possible causes and recalling the possible things they did wrong during conception and pregnancy (*n*=19/25, *Mother FDx*=5/6, *Father FDx*=5/6, *Mother PDx*=6/9, *Father PDx*=3/4). A further 6 parents attributed the condition to their own bad luck or karma (*Mother FDx*=1/6, *Father FDx*=2/6, *Mother PDx*=2/9, *Father PDx*=1/4). "Yeah, there's that question there. Was it something we did or what actually caused it? There's not many answers out there at the moment."

During this search for answers, a large proportion of mothers and fathers spoke

about blaming themselves for the condition (*n*=18/25).“For me, it was all my fault straight, you know, if [baby’s name] had something wrong it was my fault. It wasn't anything to do with [my husband]. It was my fault. He was growing inside of me.”

Problem-focused coping: Taking practical steps to address a difficult experience

Planful problem-solving

The majority of parents (*n*=23/25) reported engaging in problem-focused coping through the use of planful problem-solving, as conceptualized by Lazarus and Folkman (1984). Participants described three main ways in which planful problem-solving had occurred: attempting to accept the diagnosis and focus on what needed to be done (*n*=13/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=3/9, *Father PDx*=4/4), generally trying to get prepared and organised (*n*=12/25, *Mother FDx*= 3/6, *Father FDx*=4/6, *Mother PDx*=4/9, *Father PDx*=1/4), and breaking down the journey from diagnosis through birth and treatment into smaller, more manageable steps (*n*=18/25, *Mother FDx*=4/6, *Father FDx*=4/6, *Mother PDx*=6/9, *Father PDx*=4/4). One father discussed utilising medical staff to identify the different steps involved in their baby’s journey from start to finish; “… basically because we went from step one to end of game through everything with the doctors, everything along the way that I had a question about, I just drilled them and they answered everything.” Parents accessed multiple sources to gather information, including: the healthcare team (*n*=21), the Internet (*n*=13), and information pamphlets (*n*=2).

Confrontive coping

Mothers in both the fetal and postnatal groups reported occasions during which they engaged in confrontive coping (*n*=8/25, *Mother FDx*=4/6, *Father FDx*=0/6, *Mother PDx*=4/9, *Father PDx*=1/4). This involved standing up for what they believed in and what they wanted, such as: fighting to be connected with their baby

physically and emotionally and to take their baby home. One mother described her disregard for maternity hospital protocol in order to be with her baby as quickly and frequently soon as possible after giving birth: “But from day one I just ignored the doctor’s orders and walked down there about four times a day. So if I… I could get in trouble, but I couldn’t not be there.”

Mothers in the postnatal group described engaging in confrontive coping in order to demonstrate to others that they were indeed the expert in understanding and caring for their baby (*n*=11/25, *Mother FDx*=2/6, *Father FDx*=2/6, *Mother PDx*=5/9, *Father PDx*=2/4). Several parents, including a father in the postnatal group, recounted incidents in which their baby was crying uncontrollably and they needed to really push the medical staff in order to get some relief for the baby;

> But the most upsetting part was when the morphine was reduced and [our baby] became… , he was just frantic, screaming and carrying on and we didn't know what to do and we finally prevailed on the staff to increase the morphine.

Emotion-Focused Coping: Trying to process one’s emotions amid stress and adversity

Self-Controlling

A comparative number of mothers and fathers from the fetal and postnatal diagnostic groups described attempts to control their own emotions (*n*=7/25, *Mother FDx*=1/6, *Father FDx*=2/6, *Mother PDx*=2/9, *Father PDx*=2/4) and to keep their feelings to themselves (*n*=13/25, *Mother FDx*=3/6, *Father FDx*=4/6, *Mother PDx*=3/9, *Father PDx*=3/4). Parents spoke about controlling their own emotions in order to “stay strong” for themselves and for their partner and family. One father talked about keeping his feelings to himself to support his partner and also give himself a chance to work through his feelings on his own;

> I don't want to sort of bring anything out on [my wife]. She might have her own concerns and me voicing mine would not help her, so. Yeah, no, I just generally deal with those sorts of things by myself in my own way.

One mother in the fetal group discussed an occasion where she and her partner controlled their feelings of happiness. After surgery it was likely their baby would require a pacemaker but after several days, their baby's heart fell into a natural rhythm and it was no longer required. This mother explained that while they were happy this had occurred, they controlled their relief and happiness in order to protect themselves; "That was a massive sense of relief, but again not quite allowing ourselves to believe it. Always protecting that part of us. We'll believe it when we see it."

Distancing

All parents, with the exception of one mother in the postnatal group, engaged in some form of distancing as a method of coping. Fourteen parents described distancing their baby from the diagnosis and as a consequence actively trying to treat the baby as a "normal", healthy newborn (*n*=14/25, *Mother FDx*=5/6, *Father FDx*=2/6, *Mother PDx*=4/9, *Father PDx*=3/4). One father described distancing as an important coping mechanism that he and his family employed;

> And for us he was this, you know, part of that, you know, that coping mechanism of, you know, we were just trying to treat him as, well, we were treating him as, like a normal baby. Almost that distance from his heart condition.

Differences were identified between fetal and postnatal diagnostic groups in the way distancing was utilised. Some parents were observed to go on living their lives as if nothing had happened (*n*=11/25, *Mother FDx*=4/6, *Father FDx*=3/6, *Mother*

PDx=1/9, *Father PDx*=3/4), while parents in the postnatal group more frequently reported making light of the situation (*n*=6/25, *Mother FDx*=0/6, *Father FDx*=0/6, *Mother PDx*=2/9, *Father PDx*=4/4), and refusing to think too much about their baby's health condition (*n*=9/25, *Mother FDx*=1/6, *Father FDx*=0/6, *Mother PDx*=5/9, *Father PDx*=3/4); "So I wasn't going to spend time to explore the various possibilities or things that cannot - may or may not have developed. I think I wasn't actually very concerned. Or I tried not to become concerned too much."

Escape-Avoidance

Two main themes were identified in relation to Escape-avoidance coping. Eleven parents acknowledged that they, at times, tried to avoid being with or talking to other people (*n*=11/25, *Mother FDx*=3/6, *Father FDx*=4/6, *Mother PDx*=3/9, *Father PDx*=1/4), and a number of parents vividly described fantasies or wishes that the diagnosis would go away or that a miracle would occur and their baby's heart would be "fixed" (*n*=10/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=4/9, *Father PDx*=1/4). For example, one fetal father remembered his beliefs that his wife's morning sickness during pregnancy was her body's way of correcting their baby's heart abnormality; "I thought that that was her body working in overtime to try and correct the abnormality or the fault, either chemically or something like that."

Seeking Social Support

Sources of social support identified by parents included medical staff, partners and spouses, family, friends, and other parents of a child with CHD. A high proportion of parents, particularly mothers in the postnatal group, identified medical staff as an important source of hope, reassurance, confidence, and encouragement (Please refer to Table 8 for further differences found between the diagnostic groups) (*n*=18/25, *Mother FDx*=3/6, *Father FDx*=5/6, *Mother PDx*=8/9, *Father PDx*=2/4).

One mother recalled,

> …the nurses in ICU were amazing. They were invaluable I think in their care and commitment to the babies and just knowing that you, you know, you could walk away and know that they were completely in safe hands was very reassuring, especially knowing that they were in such a vulnerable situation.

A smaller number of parents described talking with their spouse or partner during the most stressful and difficult of times, and a smaller number of participants explicitly recalled speaking about their feelings with their partner (*n*=15/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=8/9, *Father PDx*=2/4).

Physical proximity was also identified as an important source of social support for parents. This involved physically being with their spouse or partner (*n*=11/25, *Mother FDx*=3/6, *Father FDx*=0/6, *Mother PDx*=7/9, *Father PDx*=1/4), receiving a visit in the hospital from family (*n*=10/25, *Mother FDx*=3/6, *Father FDx*=0/6, *Mother PDx*=6/9, *Father PDx*=1/4) and friends (*n*=3/25, *Mother FDx*=1/6, *Father FDx*=0/6, *Mother PDx*=2/9, *Father PDx*=0/4). Mothers in the postnatal more than the fetal group also expressed appreciation for the instrumental active support received from others, such as assistance with looking after their older children or cooking meals (*n*=14/25, *Mother FD*=3/6, *Father FD*=2/6, *Mother PD*=8/9, *Father PD*=1/4). In addition, participants also reported that reading the stories of, or speaking to, other parents of children with CHD, assisted in their own learning and coping (*n*=12/25, *Mother FDx*=2/6, *Father FDx*=2/6, *Mother PDx*=6/9, *Father PDx*=2/4). "…You know, you hear or read about kids having heart surgery and, you know it's, as much as it's fine they say, 'oh the risks are very small and are very low', and that's fine, they're still there."

Meaning-focused coping: Making meaning from great difficulty

Benefit finding

While all parents perceived their baby's diagnosis as a significant event with significant impact, a number of parents were able to find benefit in the condition and the journey they travelled with their baby as a result. Ten parents felt they had a better connection with their baby due to his or her heart condition, describing their baby as "more precious" given all they had been through together (*n* =10/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=3/9, *Father PDx*=2/4);

> I think of the other aspect of it as well. I think I certainly bonded with him far more. I think he - I really felt that he's become - he was my son. Before he was just a baby that arrived and we welcomed him. But he was actually now - he was my son. I have to look after him and… yeah.

A number of parents expressed an appreciation that their baby's heart condition was "fixable" and that other babies and their families experience worse (*n*=12/25, *Mother FDx*=4/6, *Father FDx*=1/6, *Mother PDx*=4/9, *Father PDx*=3/4). Several parents, predominantly in the postnatal diagnostic group, also acknowledged that their treatment journey with their baby was relatively short, whilst other babies and parents faced a lifelong battle (*n*=6/25, *Mother FDx*=1/6, *Father FDx*=0/6, *Mother PDx*=3/9, *Father PDx*=2/4);

> But in the same sense it was such a reality that we are so lucky that we weren't in their position, but yeah, and we didn't have the battles ahead of us that they did, I guess, which is very mean, the fact that they still had to go through a lot of stuff and we didn't. But I was quite relieved that I'd prefer to be in my position than theirs.

A higher number of parents in the postnatal group also reported feeling their

relationships with their partner and family members had been strengthened by the experience, feeling immense support and love for their relatives and others as a result of such a difficult time (*n*=10/25, *Mother FDx*= 2/6, *Father FDx*=1/6, *Mother PDx*=4/9, *Father PDx*=3/4).

Benefit reminding

A total of eight parents engaged in benefit reminding during the interview (*n*=8/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=3/9, *Father PDx*=1/4). This involved parents identifying the potential benefits of the surgery and subsequent treatment, and that their baby was getting better, had come home with them now had his or her whole future ahead of them. "It's kind of a chapter that's behind us. We're well and truly through the worst of it, so it looks like a very positive future for him. Yeah, he's just got his whole life in front of him now."

A small number of parents in the fetal group (*n*=3/12, *Mother FDx*=2/6, *Father FDx*=1/6) spontaneously talked about how lucky they felt they were to have learnt of the diagnosis in the antenatal period. These parents reported feeling grateful to have had the choice of ending their pregnancy or not, and grateful they were aware of the condition prior to taking their baby home from the obstetric ward.

Adaptive Goal Processes

A small number of parents in the fetal group reported using adaptive goal processes, such as letting go of dreams of their child being a professional athlete, when they discussed their recognition and acceptance of the limitations of their child's future (*n*=3/25, *Mother FDx*=1/6, *Father FDx* =2/6, *Mother PDx*=0/9, *Father PDx*=0/4). For example, one mother spoke of the other opportunities available for her baby in the future other than elite sport;

So we sort of look at it like, okay, well she can be an academic or a musician or

something else. If the worst that she can't be is an elite athlete, well you know me and my husband aren't anyway, so it's no big deal.

Coping processes not conceptualized within the Lazarus and Folkman model of coping: parental pride and placing focus on their baby

There was one important coping mechanisms identified in the narratives of participants that was not described in the model developed by Lazarus and Folkman. In contrast to distancing, a sizeable proportion of parents spoke about feeling proud of their baby (*n*=11/25, *Mother FDx*=3/6, *Father FDx*=2/6, *Mother PDx*=2/9, *Father PDx*=4/4), describing their baby as "fantastic" and "beautiful" (*n*=13/25, *Mother FDx*=2/6, *Father FDx*=3/6, *Mother PDx*=5/9, *Father PDx*=3/4), and several parents in the postnatal group particularly described feeling an "instant bond" with their baby (*n*=7/25, *Mother FDx*=0/6, *Father FDx*=2/6, *Mother PDx*=3/9, *Father PDx*=2/4). "But yeah very big connection there, especially when she opened her eyes and we saw each other." Mothers in the postnatal group in particular reported taking as many photos of their baby as possible, hoping to stay connected with their baby and as reassurance in case the worst happened and their baby did not survive (*n*=7/25*, Mother FDx*=0/6, *Father FDx*=1/6*. Mother PDx*=4/9, *Father PDx*=2/4); "I just wanted to spend every second with him, you know, doing everything I could, taking millions of photos, doing everything with him because you just don't know."

Discussion

In a qualitative exploration of parents' appraisals and coping responses to a diagnosis of complex CHD in their baby, this article strived to accomplish three main aims. The first was to determine if Lazarus and Folkman's model of stress and coping (1984) was relevant and applicable to the paediatric medical setting. While it

was found that the majority of coping strategies implemented by participants fell within the structure of the model, parental pride and placing focus on their baby could not be adequately classified. This coping strategy has previously been identified in the literature. For example, in a study exploring coping in parents who had a baby in the Neonatal Intensive Care Unit, focusing on the newborn was identified as one of the primary coping strategies employed (Hughes, McCollum, Sheftel & Sanchez, 1994). This finding highlights a limitation in the model posited by Lazarus and Folkman, in terms of capturing the reciprocal nature of relationships, and the strength and assistance that can be drawn from those relationships.

The second aim of the article was to identify and compare appraisals and coping strategies utilised by parents who received a fetal diagnosis with parents who received a postnatal diagnosis. Several significant thematic differences were found between the two diagnostic groups. First, a larger number of parents who received a fetal diagnosis reported “feeling prepared” compared with parents who received a postnatal diagnosis, the latter of whom were more likely to report feeling “helpless” and “out of control”. This finding lends support to previous studies that found a fetal diagnosis is perceived by most parents as providing the opportunity to prepare and plan for the arrival of their medically fragile baby (Hoehn et al., 2004). Yet this finding needs to be questioned as further analysis found a sizable proportion of fathers in the fetal diagnostic group also reported feeling as though they didn’t know what was going to happen or what they would do. We propose that it is difficult for mothers and fathers to feel prepared to parent a child with a complex CHD diagnosis, regardless of the time of diagnosis. Research suggests that even the task of preparing a parent to care for their infant after surgery is an arduous one and medical staff have

an important role to play in providing the appropriate information and support to achieve a level of preparedness in parents (Sherry & Green, 2003).

Consistent with the model, all but one parent engaged in some form of distancing as a means of coping. In particular, a large number of mothers who received a fetal diagnosis and fathers in both the fetal and postnatal diagnostic groups reported trying to carry on as if nothing happened. In contrast, the majority of mothers who received a postnatal diagnosis did not report engaging in this form of distancing.

Reasons for this finding are unclear, although it is speculated mothers and fathers who received a fetal diagnosis reported engaging in this form of distancing, both before and after surgery, in attempts to distance their baby from the CHD diagnosis. Parents who receive a fetal diagnosis are not given the opportunity to meet and get to know their baby without the knowledge of their condition. Turning to the literature regarding a different condition, namely cystic fibrosis, it has been found that once a parent learns of their baby's diagnosis, their "sense of who the child is" has changed and is enmeshed with the condition diagnosed (Grob, 2008, p.1063). Rather than getting to know and love their baby, the parent's focus is instead centred on the condition and it's treatment (Grob, 2008). It is important to note that placing focus on the baby was a significant theme identified within a large number of parent narratives. The type of distancing described in the present study may be interpreted as a way to refocus parent's attention back onto the baby and has previously been identified as a prominent coping strategy in the literature, labelled as normalisation (Lee & Rempel, 2011). A study undertaken in 2011 found that normalisation was a coping strategy employed by parents of infants with hypoplastic left heart syndrome

as a way to fight to recognise their infant as a ‘normal’ baby not defined by his or her condition (Lee & Rempel).

It is further speculated that the differences found between mothers and fathers in the postnatal diagnostic group may be due to the variations in reported primary and secondary appraisals of the condition. Mothers who received a postnatal diagnosis were more inclined than parents in the three other diagnostic groups to use expressive and descriptive language when describing the impact of the diagnosis, labelling it as life-changing and stating it caused their world to fall apart. This appraisal directly conflicts with this form of distancing, it would be very difficult and ineffective for mothers who received a postnatal diagnosis to simultaneously appraise the situation in this manner whilst also trying to carry on as if nothing had happened.

While parents in both the fetal and postnatal diagnostic groups engaged in meaning-focused coping, the type with which they employed differed. Several parents who received a fetal diagnosis engaged in adaptive goal processes, effectively recognising and accepting the limitations of their child’s future, while none of the parents who received a postnatal diagnosis discussed this concept. Based on this, it is hypothesised that parents who received a fetal diagnosis were better equipped to engage in adaptive goal processes because they learned about the diagnosis prior to birth and as such, the diagnosis was already part of the identity of their baby (Grob, 2008). As a consequence, it is suggested that parents who received a fetal diagnosis were more readily able to accept the limitations placed upon their infant and to acknowledge and accept the long-term implications. Other practical reasons could contribute to this finding, including parents in the fetal diagnostic group having greater opportunity to speak with medical staff regarding their baby’s future and the long-term consequences of the condition.

A much larger proportion of parents in the postnatal group engaged in benefit finding and reported their relationship with their partner and family was strengthened as a result of the stressful experience. Unlike parents who received a fetal diagnosis, parents in the postnatal diagnostic group had the opportunity to form an identity of their baby as being healthy and separate to the condition, if only for a very short time. Consequently, it is hypothesed that parents who receive a postnatal diagnosis find meaning in their experiences in a different way, instead trying to find a way to reconcile with the loss of their 'healthy' baby but also fighting strongly to keep their baby separate from their diagnosis.

A further consideration upon receiving a postnatal diagnosis in their baby, parents are often required to act quickly and to address the condition, at times, within hours. It is theorised that parents in the postnatal diagnostic group reported the condition strengthened their relationships due to these significant pressures. Parents in the fetal diagnostic group, whilst also under immense pressure, were given more time to plan and prepare for their baby's treatment and therefore may not have needed to rely so heavily on their relationships for support (Hoehn et al., 2004).

Diagnostic groups also differed in their reported sources of support and reassurance. Several parents who received a fetal diagnosis spoke about faith as a source of reassurance. In contrast, a large group of parents who received a postnatal diagnosis sought reassurance from several other sources. For instance, a higher proportion of parents who received a postnatal diagnosis spoke about having an instant bond with their baby and feeling connected to their baby from birth. Further, a number of parents in the postnatal diagnostic group also spoke more about taking photos of their baby. Several of these parents described these photos as precious and important, particularly given the uncertainty surrounding their baby's future and the

possibility of death. The process of attachment between a parent and their baby begins in the fetal period (Franklin, 2006) and as discussed above, parents who have received a fetal diagnosis have previously reported finding their attachment has been disrupted as their focus has shifted solely onto the condition itself (Grob, 2008). Parents who receive a postnatal diagnosis, on the other hand, are likely still afforded the opportunity to bond and connect with their baby during pregnancy (Franklin, 2006; Grob, 2008). We suggest that parents who receive a fetal diagnosis may be unable to form as strong a bond as reported by parents who receive a postnatal diagnosis due to this focus on the condition itself and this interruption in attachment during the pregnancy.

The third aim of the study was to explore potential similarities and differences between mothers and fathers in their experience of receiving a diagnosis of CHD in their baby. Consistent with the model of stress and coping, all parents engaged in a primary appraisal of the diagnosis, perceiving it as a significant and important event, irrespective of gender or timing of diagnosis.

Interestingly, similar proportions of mothers and fathers reported engaging in problem-focused coping across all four groups. While it has been hypothesised in the literature that men are more inclined to utilise problem-focused coping than women, recent findings have found that women engage in problem-focused coping as frequently (Maltaud, 2004; Melendez et al., 2012) or more often than men (Tamres, Janicki and Helgeson, 2002). This result suggests that problem-focused coping is just as important for mothers as it is for fathers, and in the present study, that the acquisition of health-related information is an important means of coping for both mothers and fathers.

Mothers were found, however, to differ from fathers in several ways. For instance, there was wide variation in emotional responses to diagnosis and while the majority of mothers reported feeling “shocked” and “scared”, these emotions were rarely reported by fathers. Mothers who had received a postnatal diagnosis were especially expressive in describing the magnitude of the diagnosis and the impact it had upon their lives. Consequently, our data suggests that mothers appraised receiving a diagnosis of complex CHD in their baby as more stressful and significant than fathers. Previous research lends support to this finding. When presented with the same scenario, women have been found to rate the scenario as more stressful than men (Eaton & Bradley, 2008). It is possible that this identified disparity may be indicative of a difference in experience; however, it could also reflect differences in the nature or level of reporting between mothers and fathers.

While mothers and fathers both engaged in confrontive coping, mothers were more inclined to utilise certain forms of confrontive coping than fathers. Mothers more frequently reported making demands to be physically close to their baby. This involved fighting to get to their baby, to take their baby home, or to stop medical staff from taking their baby away for surgery. There may be several reasons for this finding. A plausible explanation is that mothers needed to utilise confrontive coping more frequently than fathers due to restrictions placed upon them by medical staff soon after birth. Mothers, at times, had limited access to their baby directly after birth due to the baby being transferred to either another hospital or special care unit, whilst the mothers needed to remain in the maternity ward.

A further consideration is that this could be linked with the possibility that mothers place greater importance on trying to regain control of the care of their baby (Jackson, Ternestedt & Schollin, 2003). In a study of parents of premature babies,

mothers reported a need to gain control and to get involved with their baby's care more frequently than fathers (Jackson et al., 2003). In the same study, fathers reported restrictions in their ability to be at the hospital with their baby due to other commitments (such as work) and therefore, placed greater faith and confidence in medical staff to look after their baby (Jackson et al., 2003). These findings suggest that while mothers try to gain control of the care of their baby in the hospital, fathers instead place their faith in the medical staff to care for and protect their baby.

The current study does not lend support to the above argument as a proportion of fathers described engaging in another form of confrontive coping, namely, fighting for others to recognise they know how to best look after their baby and to identify when something is wrong. While fathers did not discuss fighting to be physically close to their baby, a group of fathers did highlight their battle to be heard by medical staff in order to access the best medical care for their baby. This finding suggests that fathers are not willing to only place their faith in the medical staff and they are also willing to step up and to fight for the best care for their baby.

Consistent with the literature, mothers were observed to utilise emotion-focused coping more frequently than fathers, specifically in their engagement in seeking social support from family, friends and their spouses or partners (Eaton & Bradley, 2008). Interestingly, a greater proportion of mothers in the postnatal diagnostic group, compared to the three other groups, reported talking to their partners about their feelings. One possible explanation for this finding may be due to the fact that mothers who receive a postnatal diagnosis have less time to process the emotions that are elicited by the diagnosis and treatment of the condition (Rychik et al., 2013). Access to social supports are likely limited as mothers are confined to the

hospital and, as a consequence, need to draw upon resources available to them such as their spouses or partners.

Mothers also appeared to differ from fathers in the meaning they took from their experience with their sick baby. A larger proportion of mothers engaged in benefit finding, expressing an appreciation of their baby's condition as treatable and recognising that their baby's condition could have been worse. This finding may simply reflect the reality experienced by mothers, their baby has undergone treatment, has predominantly recovered and they are now able to look back on their baby's journey as a large hurdle that was overcome. Another possible explanation for this finding may be linked to the previously mentioned notion that mothers experienced a strong need to regain control of the situation when faced with their baby's medical condition and required treatment when compared to fathers (Jackson et al., 2003). It is theorised that mothers who perceived their baby's condition as fixed, short term and could have been much worse is a way for mothers to take back this control and to prevent the diagnosed condition from having as strong an influence on their baby's life (Jackson et al., 2003).

Study Limitations

Several limitations of this study need to be highlighted. First, the small sample utilised places restrictions on the generalizability of the findings and the qualitative nature of the study prevents the formulation of statistically valid generalisations. It is suggested that the use of a larger sample size would assist in gathering stronger support for thematic patterns identified in our analysis and a quantitative analysis would provide an opportunity to form statistically valid generalisations and to identify statistically significant differences between parents based on time of diagnosis or gender.

Second, while the study was able to explore the role of gender and compare mothers with fathers, an exploration of the impact of parent age was not within the scope of the current study. It has been suggested that age can influence the use of coping strategies, with several studies finding younger adults are more inclined to appraise a stressful situation as changeable and to utilise active, problem-focused coping, while older adults tend to appraise stressful situations as unchangeable and, as a consequence, utilised more emotion-focused coping (Folkman, Lazarus, Pimlet and Novacek, 1987). Subsequently, differences in the age of parents may account for differences in coping strategies utilised rather than gender or time of diagnosis and as such is an interesting and potentially important variable to consider in future studies in relation to this population.

Third, the time from diagnosis to interview was significantly different between the diagnostic groups, with parents who received a fetal diagnosis having a significantly longer time from diagnosis to interview when compared to parents who received a postnatal diagnosis. This difference is difficult to avoid due to the nature of the study and the comparison between the two diagnostic groups. Regrettably, differences found between diagnostic groups may be a result of this significant difference in time since diagnosis, rather than in experience and impact of timing of diagnosis. Parents in the fetal diagnostic group may have been further along in their journey with their baby to those parents who received a postnatal diagnosis and therefore naturally engaged in different coping strategies as a result. It is recommended that further research be designed and conducted in a fashion that tries to control for the time of interview since diagnosis, thereby making the two diagnostic groups more comparable.

Finally, it was also beyond the scope of the current study to consider the role of hope in mothers and fathers who receive a diagnosis of CHD in their baby. The dynamic and dependent relationship between coping and hope has recently been identified in the literature (Folkman, 2010) and as such, the investigation of the role of hope in this population is theorised to strengthen our understanding of parents coping responses during times of stress. Currently, the literature provides forty-nine different definitions for hope (Schrank, Stanghellini & Slade, 2008). In order to examine hope within this area, a definition must be attained and this was considered beyond the scope of the current study.

Clinical and research implications and recommendations

The findings of the present article suggest that Lazarus and Folkman's model of stress and coping appears to be helpful in understanding and categorising the basic coping strategies utilised by parents who have received a diagnosis of complex CHD in their baby. However, the model appears limited in terms of facilitating an appreciation and recognition of the importance of parental pride and focus on their baby. This result is beneficial in providing guidance for clinicians working with this population in the types of coping utilised as well as the need to place importance on the relationship between the parent and their baby as a source of coping and support. It needs to be a key concern for medical staff to facilitate and encourage attachment between the infant and their parents particularly as hospitalisation has been reported to disrupt this natural event (Franklin, 2006). Specifically considering our findings, this support seems crucial for parents who received a fetal diagnosis as their attachment appears to have also been disrupted by the diagnosis during pregnancy (Franklin, 2006; Grob, 2008).

The present study did not provide conclusive evidence to indicate that timing of diagnosis has a significant influence on the psychological and emotional outcomes of parents after receiving a diagnosis of complex CHD in the infant. It did, however, highlight the need for medical staff to try to help parents, particularly those in the postnatal diagnostic group, to feel better prepared for the treatment of their baby. This will likely include assisting parents in utilising problem-focused coping such as information gathering and speaking with doctors. Medical staff are also encouraged to help parents, particularly those in the fetal diagnostic group, in separating their baby from the diagnosis. Previous research has made the suggestion that this can be achieved by explicitly asking parents how much information they would like about the condition, the treatment and the prognosis (Grob, 2008). While this may not be as applicable in relation to such a life-threatening condition as complex CHD, it is suggested that future research could investigate further the level of information that parents would like to receive regarding their baby's diagnosis and prognosis and the mode in which they would like to receive it.

In highlighting differences between mothers and fathers in this population, the current study has highlighted the importance of emotion-focused coping, particularly in mothers. Medical staff can help to facilitate the use of this type of coping by providing opportunities for mothers to discuss and express their emotions and to encourage mothers to access social supports around them.

In terms of further research, our findings suggest that different forms of meaning-focused coping are adopted by parents across diagnostic groups and when comparing mothers with fathers. Differences were also identified in sources of reassurance between fetal and postnatal diagnostic groups. After comparing mothers and fathers we found that problem-focused coping was widely used by both but

mothers more frequently reported applying emotion-focused coping. Currently, there is no gold standard for the measurement of coping in the literature (Folkman & Moskowitz, 2004). It has been suggested that the approach we utilised in our analysis is the preferred method of preliminary exploration of coping in a specific population as this approach then identifies stressors and coping strategies frequently employed that can then be targeted in a quantitative exploration (Folkman & Moskowitz, 2004). It is therefore recommended that further research explore and expand the findings of our analysis by engaging a larger sample size and utilising quantitative measures and specifically target parents use of meaning-focused coping, emotion-focused coping and sources of reassurance.

Also of interest are the limitations found in Lazarus and Folkman's model of stress and coping. The model was unable to adequately measure and categorise the parent/infant relationship as an important source of support and a way for parents to cope. We further recommend future research endeavour to expand the model to encapsulate this relationship and adequately recognise the importance of attachment for parents as they try to cope with their baby's medical condition.

Conclusions

When faced with a diagnosis of complex CHD in their baby, parents must find a way to cope. This study found that Lazarus and Folkman's model of stress and coping is predominantly effective in categorising the main types of coping employed by parents who have received such a diagnosis; however, it fails to appreciate the power and strength drawn from the relationship between parents and their baby. This study also found that overall, it was difficult to be prepared to be a parent to a baby with complex CHD and many parents attempted (consciously or unconsciously) to

cope by distancing and separating their baby from the medical condition. The findings suggest that the meaning parents found in, or made from, their experiences and the ways in which they sought reassurance differed between groups. Mothers were found to utilise emotion-focused coping more frequently than fathers and were readily able to find benefit and meaning from their journey with their sick baby. Consequently, this analysis has demonstrated important avenues along which further research may journey in order to better understand the experiences and needs of parents of a baby with a diagnosis of complex CHD and ways in which they can be supported as they try to cope.

References

AIHW 2011. Cardiovascular disease: Australian facts 2011. Cardiovascular disease series no. 35. Cat. no. CVD 53. Canberra: AIHW. Viewed 18 May 2014 http://www.aihw.gov.au/publication-detail/?id=10737418510 p 127.

Bazeley, P. (2013). *Qualitative data analysis: Practical strategies*. London: Sage.

Beutler, L. E., Moos, R. H., & Lane, G. (2003). Coping, treatment planning, and treatment outcome: discussion. *Journal of Clinical Psychology*, *59*(10), 1151-1167. doi: 10.1002/jclp.10216

Blue, G. M., Kirk, E. P., Sholler, G. F., Harvey, R. P., & Winlaw, D. S. (2012). Congenital heart disease: Current knowledge about causes and inheritance. *The Medical Journal of Australia*, *197*(3), 155-159.

Boyd, J. D., & Hamilton, W. J. (1967). Foetus—or Fetus? *British Medical Journal*, *1*(5537), 425.

Brosig, C. L., Whitstone, B. N., Frommelt, M. A., Frisbee, S. J., & Leuthner, S. R. (2007). Psychological distress in parents of children with severe congenital heart disease: The impact of prenatal versus postnatal diagnosis. *Journal of Perinatology*, *27*(11), 687-692. doi:10.1038/sj.jp.7211807

Clark, S. M., & Miles, M. S. (1999). Conflicting responses: The experiences of fathers of infants diagnosed with severe congenital heart disease. *Journal for Specialists in Pediatric Nursing*, *4*(1), 7-14. doi: 10.1111/j.1744-6155.1999.tb00075.x

Davis, C. C., Brown, R. T., Bakeman, R., & Campbell, R. (1998). Psychological adaptation and adjustment of mothers of children with congenital heart disease: Stress, coping, and family functioning. *Journal of Pediatric Psychology*, *23*(4), 219-228. doi: 10.1093/jpepsy/23.4.219

Doherty, N., McCusker, C. G., Molloy, B., Mulholland, C., Rooney, N., Craig, B., ... & Casey, F. (2009). Predictors of psychological functioning in mothers and fathers of infants born with severe congenital heart disease. *Journal of Reproductive and Infant Psychology*, *27*(4), 390-400. doi:10.1080/02646830903190920

Eaton, R. J., & Bradley, G. (2008). The role of gender and negative affectivity in stressor appraisal and coping selection. *International Journal of Stress Management*, *15*(1), 94-115. doi: 10.1037/1072-5245.15.1.94

Folkman, S. (2010). Stress, coping, and hope. *Psycho-Oncology, 19*, 901-908. doi: 10.1002/pon.1836

Folkman, S. (2008). The case for positive emotions in the stress process. *Anxiety, Stress, and Coping*, *21*(1), 3-14. doi:10.1080/10615800701740457

Folkman, S., Lazarus, R. S., Dunkel-Schetter, C., DeLongis, A., & Gruen, R. J. (1986). Dynamics of a stressful encounter: Cognitive appraisal, coping, and encounter outcomes. *Journal of Personality and Social Psychology*, *50*(5), 992. doi: http://dx.doi.org/10.1037/0022-3514.50.5.992

Folkman, S., Lazarus, R. S., Pimley, S., & Novacek, J. (1987). Age differences in stress and coping processes. *Psychology and Aging*, *2*(2), 171-184. doi: http://dx.doi.org/10.1037/0882-7974.2.2.171

Folkman, S., & Moskowitz, J. T. (2000). Positive affect and the other side of coping. *American Psychologist*, *55*(6), 647-654. doi: http://dx.doi.org/10.1037/0003-066X.55.6.647

Folkman, S., & Moskowitz, J. T. (2004). Coping: Pitfalls and promise. *Annual Review of Psychology*, *55*, 745-774. doi: 10.1146/annurev.psych.55.090902.141456

Fonseca, A., Nazaré, B., & Canavarro, M. C. (2012). Parental psychological distress and quality of life after a prenatal or postnatal diagnosis of congenital anomaly: A controlled comparison study with parents of healthy infants. *Disability and Health Journal*, *5*(2), 67-74. doi:10.1016/j.dhjo.2011.11.001

Franklin, C. (2006). The neonatal nurse's role in parental attachment in the NICU. *Critical Care Nursing Quarterly*, *29*(1), 81-85.

Frydenberg, E. (2014). Coping research: Historical background, links with emotion, and new research directions on adaptive processes. *Australian Journal of Psychology*, *66*(2), 82-92. doi: 10.1111/ajpy.12051

Gilgun, J. F. (1995). We shared something special: The moral discourse of incest perpetrators. *Journal of Marriage and the Family*, *5*(2), 265-281.

Grob, R. (2008). Is my sick child healthy? Is my healthy child sick?: Changing parental experiences of cystic fibrosis in the age of expanded newborn screening. *Social Science & Medicine*, *67*(7), 1056-1064, p. 1063. doi: doi:10.1016/j.socscimed.2008.06.003

Haan, N. (1965). Coping and defense mechanisms related to personality inventories. *Journal of Consulting Psychology*, *29*(4), 373-378. doi: http://dx.doi.org/10.1037/h0022410

Harvey, K. A., Kovalesky, A., Woods, R. K., & Loan, L. A. (2013). Experiences of mothers of infants with congenital heart disease before, during, and after complex cardiac surgery. *Heart & Lung: The Journal of Acute and Critical Care*, *42*(6), 399-406. doi: http://dx.doi.org/10.1016/j.hrtlng.2013.08.009

Hobfoll, S. E. (1989). Conservation of resources: A new attempt at conceptualizing stress. *American Psychologist*, *44*(3), 513. doi: http://dx.doi.org/10.1037/0003-066X.44.3.513

Hobfoll, S. E. (2001). The influence of culture, community, and the nested‐self in the stress process: Advancing conservation of resources theory. *Applied Psychology*, *50*(3), 337-421. doi: 10.1111/1464-0597.00062

Hoehn, K. S., Wernovsky, G., Rychik, J., Tian, Z. Y., Donaghue, D., Alderfer, M. A., ... & Nelson, R. M. (2004). Parental decision-making in congenital heart disease. *Cardiology in the Young*, *14*(03), 309-314. doi: http://dx.doi.org/10.1017/S1047951104003099

Hughes, M. A., McCollum, J., Sheftel, D., & Sanchez, G. (1994). How parents cope with the experience of neonatal intensive care. *Children's Health Care*, *23*(1), 1-14. doi: 10.1207/s15326888chc2301_1

Jackson, K., Ternestedt, B. M., & Schollin, J. (2003). From alienation to familiarity: Experiences of mothers and fathers of preterm infants. *Journal of Advanced Nursing*, *43*(2), 120-129. doi: 10.1046/j.1365-2648.2003.02686.x

Kasparian N. A**.**, Deans, C.E., Kirk, E. P., Barnett, B. E., Winlaw, D. S., Sholler, G. F. When hope is lost and found again: Parents' experiences of fetal or early postnatal diagnosis of complex congenital heart disease. In preparation.

Lawoko, S., & Soares, J. J. (2002). Distress and hopelessness among parents of children with congenital heart disease, parents of children with other diseases, and parents of healthy children. *Journal of Psychosomatic Research*, *52*(4), 193-208. doi: doi:10.1016/S0022-3999(02)00301-X

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

Lee, A., & Rempel, G. R. (2011). Parenting children with hypoplastic left heart syndrome: Finding a balance. *Journal for Specialists in Pediatric Nursing*, *16*(3), 179-189. doi: 10.1111/j.1744-6155.2011.00289.x

Livneh, H., & Martz, E. (2007). An introduction to coping theory and research. In *Coping with chronic illness and disability* (pp. 3-27). Springer US.

Matud, M. P. (2004). Gender differences in stress and coping styles. *Personality and Individual Differences*, *37*(7), 1401-1415. doi: doi:10.1016/j.paid.2004.01.010

Meléndez, J. C., Mayordomo, T., Sancho, P., & Tomás, J. M. (2012). Coping strategies: Gender differences and development throughout life span. *The Spanish Journal of Psychology*, *15*(03), 1089-1098. doi: http://dx.doi.org/10.5209/rev_SJOP.2012.v15.n3.39399

Menahem, S., & Grimwade, J. (2005). Pre-natal counselling—helping couples make decisions following the diagnosis of severe heart disease. *Early Human Development*, *81*(7), 601-607. doi:10.1016/j.earlhumdev.2005.02.001

Miles, M., & Huberman, A. (1994). *Qualitative Data Analysis: An Expanded Sourcebook*. London: Sage.

Mirowsky, J., & Ross, C. E. (1995). Sex differences in distress: Real or artifact?. *American Sociological Review*, *6*(3), 449-468.

Pancer, S. M., Pratt, M., Hunsberger, B., & Gallant, M. (2000). Thinking ahead: Complexity of expectations and the transition to parenthood. *Journal of Personality*, *68*(2), 253-279. doi: 10.1111/1467-6494.00097

Pearlin, L. I., & Schooler, C. (1978). The structure of coping. *Journal of Health and Social Behavior*, 2-21.

Pye, S., & Green, A. (2003). Parent education after newborn congenital heart surgery. *Advances in Neonatal Care*, *3*(3), 147-156. doi: 10.1016/S1536-0903(03)00075-4

Quine, L., & Pahl, J. (1991). Stress and coping in mothers caring for a child with severe learning difficulties: A test of Lazarus' transactional model of coping.

Journal of Community & Applied Social Psychology, *1*(1), 57-70. doi: 10.1002/casp.2450010109

Rychik, J., Donaghue, D. D., Levy, S., Fajardo, C., Combs, J., Zhang, X., ... & Diamond, G. S. (2013). Maternal psychological stress after prenatal diagnosis of congenital heart disease. *The Journal of Pediatrics*, *162*(2), 302-307. doi:10.1016/j.jpeds.2012.07.023

Schrank, B., Stanghellini, G., & Slade, M. (2008). Hope in psychiatry: A review of the literature. *Acta Psychiatrica Scandinavica*, *118*(6), 421-433. doi: 10.1111/j.1600-0447.2008.01271.x

Sholler, G. F., Kasparian, N. A., Pye, V. E., Cole, A. D., & Winlaw, D. S. (2011). Fetal and post‐natal diagnosis of major congenital heart disease: Implications for medical and psychological care in the current era. *Journal of Paediatrics and Child Health*, *47*(10), 717-722. doi: 10.1111/j.1440-1754.2011.02039.x.

Sira, N., Desai, P. P., Sullivan, K. J., & Hannon, D. W. (2014). Coping Strategies in Mothers of Children with Heart Defects: A Closer Look Into Spirituality and Internet Utilization. *Journal of Social Service Research*, *40*(5), 606-622. doi: 10.1080/01488376.2014.908808

Skari, H., Malt, U. F., Bjornland, K., Egeland, T., Haugen, G., Skreden, M., ... & Emblem, R. (2006). Prenatal diagnosis of congenital malformations and parental psychological distress—a prospective longitudinal cohort study. *Prenatal Diagnosis*, *26*(11), 1001-1009. doi: 10.1002/pd.1542

Soulvie, M. A., Desai, P. P., White, C. P., & Sullivan, B. N. (2012). Psychological distress experienced by parents of young children with congenital heart defects: A comprehensive review of literature. *Journal of Social Service Research*, *38*(4), 484-502. doi: 10.1080/01488376.2012.696410

Spijkerboer, A. W., Helbing, W. A., Bogers, A. J., Van Domburg, R. T., Verhulst, F. C., & Utens, E. M. (2007). Long-term psychological distress, and styles of coping, in parents of children and adolescents who underwent invasive treatment for congenital cardiac disease. *Cardiology in the Young*, *17*(06), 638-645. doi: http://dx.doi.org/10.1017/S1047951107001333

Svavarsdottir, E. K., & McCubbin, M. (1996). Parenthood transition for parents of an infant diagnosed with a congenital heart condition. *Journal of Pediatric Nursing*, *11*(4), 207-216. doi: 10.1016/S0882-5963(96)80093-5

Tak, Y. R., & McCubbin, M. (2002). Family stress, perceived social support and coping following the diagnosis of a child's congenital heart disease. *Journal of Advanced Nursing*, *39*(2), 190-198. doi: 10.1046/j.1365-2648.2002.02259.x

Tamres, L. K., Janicki, D., & Helgeson, V. S. (2002). Sex differences in coping behavior: A meta-analytic review and an examination of relative coping. *Personality and Social Psychology Review*, *6*(1), 2-30. doi:10.1207/S15327957PSPR0601_1

Teti, M., Pichon, L., Kabel, A., Farnan, R., & Binson, D. (2013). Taking pictures to take control: Photovoice as a tool to facilitate empowerment among poor and racial/ethnic minority women with HIV. *Journal of the Association of Nurses in AIDS Care*, *24*(6), 539-553. doi: http://dx.doi.org/10.1016/j.jana.2013.05.001

Tracey, N. E. (2000). *Parents of premature infants: Their emotional world*. United States: Whurr Publishers.

Zaumseil, M., & Schwarz, S. (2014). Understandings of coping: A critical review of coping theories for disaster contexts. In *Cultural psychology of coping with disasters* (pp. 45-83). Springer New York.

Table 3. Conceptually Clustered Table reflecting themes expressed by participants

Theme	Number of participants who discussed this theme (*N*=25)	Fetal group		Postnatal group	
		Mothers (*N*=6)	Fathers (*N*=6)	Mothers (*N*=9)	Fathers (*N*=4)
Primary Appraisal					
This diagnosis is significant to me	25	6	6	9	4
Caused parent to cry	9	5	1	3	0
Parents worried about their child, worried about them having the surgery, worried they won't survive	21	5	6	7	3
World fell apart', 'biggest thing ever', pretty big bombshell'	12	2	1	7	2
How can this be happening to us?	9	3	3	2	1
My own bad luck, karma	6	1	2	2	1
It's not fair' - 'I just want it healthy'	3	1	1	1	0
Emotional Response to diagnosis					
Shock	12	5	1	5	1
Devastated	6	3	0	1	2
Scared	10	3	1	5	1
It felt surreal	8	1	3	3	1
Secondary appraisal – I have the resources that mean I can handle this					
Parents generally felt prepared	8	3	3	2	0
Parents felt they were able to *'deal with it'*	10	2	3	3	2
Parent reports having a supportive partner	4	2	0	1	1
Parent now has information and know what is going to happen	6	1	1	1	3
Parent feels they have access to the best medical care available	8	2	4	0	2
Secondary appraisal – I'm not sure how I am going to cope					
Didn't know what was going to happen, what they were going to do	8	1	3	4	0
Feel helpless, out of control, can't help your baby	8	0	2	3	3
Don't have the resources available (e.g., time, people for support, money, energy)	5	1	1	2	1
You just cannot prepare for it	2	1	1	0	0
Didn't deal with it	3	1	0	2	0
Confrontive coping					
Fighting for connection with the baby to physically be with the baby, to take them home and to stop others from taking baby from them	8	4	0	3	1
Parent believes they know best, they know how to look after their baby and when something is wrong	11	2	2	5	2
Self-Controlling coping					
Kept others from knowing how bad things were and tried to keep things to myself	13	3	4	3	3
Parents convinced themselves to be strong, to control their feelings	7	1	2	2	2
Parents controlled their feelings of happiness when something goes well for the baby	1	1	0	0	0
Accepting Responsibility					
Criticising and lecturing self	8	3	2	3	0
Blamed self	18	5	5	6	2
Looking for a reason/questioning	19	5	5	6	3
Planful problem solving					
Accepting the diagnosis and focusing on what needs to be done	13	2	4	3	4
Get prepared and organised in general	12	3	4	4	1
Breaking down the journey into small steps	18	4	4	6	4
Organising life around the operation	8	1	3	3	1
Distancing					
Distanced baby from the diagnosis (Newborn is a baby with a condition, not defined by the condition)	14	5	2	4	3
Parent believing and convincing themselves that they aren't feeling anything 'I'm fine'	10	1	4	3	2
Symptoms in the baby aren't that bad (the baby is asymptomatic)	5	1	2	1	1
Went along with fate, sometimes I just have bad luck	6	0	3	2	1

Theme	**Number of participants who discussed this theme** (*N*=25)	Fetal group		Postnatal group	
		Mothers (*N*=6)	Fathers (*N*=6)	Mothers (*N*=9)	Fathers (*N*=4)
Distancing					
Tried to forget the whole thing after the surgery	3	0	1	1	1
Didn't let it get to me; refused to think about it too much	9	1	0	5	3
Went on as if nothing happened	11	4	3	1	3
Made light of the situation; refused to get too serious	6	0	0	2	4
Parent distanced themselves from the baby through statistics, names, not opening baby things	8	1	3	2	2
Parent doesn't think/believe/consider that the diagnosis could be serious - prior to finding out the specific diagnosis	4	0	2	1	1
Escape avoidance					
Avoided being with/talking to people	11	3	4	3	1
Had fantasies/wishes about the outcome, wished situation would go away, hoped for a miracle	10	2	3	4	1
Took it out on other people	4	0	1	2	1
Seeking social support – Medical staff					
Talked to someone to find out more about the situation/ found someone that could do something more concrete about the problem	24	5	6	9	4
Medical staff provided hope/reassurance/confidence/ encouragement	18	3	5	8	2
Medical staff helped parents to get more information	10	4	1	3	2
Parents given advice by medical staff	11	3	2	4	2
Parents who felt that talking to Nadine was Nadine was really helpful	7	1	3	0	3
Seeking social support - Partner					
Parents talked to their partner about how they were feeling	15	2	3	8	2
Parent referred to talking to their partner	17	4	3	8	2
Parent spoke about being physically with their partner	11	3	0	7	1
Seeking social support - Family					
Notifying family members about the condition	8	1	2	3	2
Talked to someone about how I was feeling	8	3	1	3	1
Family physically came to the hospital to visit	10	3	0	6	1
Family members provided active support	14	3	2	8	1
Parents took comfort from their other children being there	4	0	0	3	1
It felt like the baby had become part of the family	3	0	0	2	1
Seeking social support - Friends					
Friends physically came to the hospital to visit	3	1	0	2	0
Friends provided active support	2	0	0	2	0
Parents were willing to tell friends, work colleagues about the condition	9	1	2	4	2
Meaning-focused coping – Benefit finding					
Appreciation that condition is 'fixable' (things could be much worse)	9	4	1	3	1
Baby is more 'precious' than a normal baby (better connection with the baby)	10	2	3	3	2
Strengthened relationships with family/partner	10	2	1	4	3
Meaning-focused coping - Benefit reminding					
Baby can and is getting better and 'we got to bring him home'	8	2	3	2	1
Knew the baby had to have the operation to get better	6	1	2	2	1
Lucky to have learnt of the diagnosis in the fetal period	3	2	1	0	0
Meaning-focused coping – Adaptive goal processes					
Parents recognise and accept the limitations of the child's future	3	1	2	0	0
Reordering Priorities					
All energy and attention went on the baby	8	0	2	4	2
Infusing ordinary events with positive meaning					
Smiling/laughing/giggling	10	2	3	3	2
Bath time	8	1	1	3	3
Breastfeeding/Feed times	4	1	0	2	1
Proximity					
Parent wants to be physically close to or holding their baby	20	6	4	9	1

Table 4. Interview Characteristics (N=25)

		Total Sample	Fetal Cardiac Diagnosis		Postnatal Cardiac Diagnosis	
Variable	**Level**	Parents (*N*=25)	Mothers (*n*=6)	Fathers (*n*=6)	Mothers (*n*=9)	Fathers (*n*=4)
Interview venue	Parent's home	5	2	2	1	0
	Paediatric hospital	4	2	2	0	0
	Telephone	16	2	2	8	4
Interview format	Couple	6	3	3	0	0
	Individual	19	3	3	9	4
Time of interview	Prior to surgery	2	1	1	0	0
	After surgery	17	2	2	9	4
	Prior to 2nd surgery	4	2	2	0	0
	After 2nd surgery	2	1	1	0	0
Baby in hospital at time of interview	Yes	3	1	2	0	0
	No	22	5	4	9	4

		Total Sample	Fetal Cardiac Diagnosis		Postnatal Cardiac Diagnosis	
Variable		Parents	Mothers	Fathers	Mothers	Fathers
Time since cardiac diagnosis (weeks)	Mean (SD)	20.1 (10.7)	26.0 (12)	27.3 (14)	12.9 (3.6)	16.1 (2.2)
	Median	16.1	22	22.2	13.7	15.1
	Range	8-47	14-41	14-47	8.17	15-19
Interview length (minutes)	Mean (SD)	85.5 (24.7)	82.9 (26.6)	80.9 (26.3)	82.8 (27.1)	102.2 (12.0)
	Range	49-126	49-124	61-124	53-126	88-113
Age of baby at interview (weeks)	Mean (SD)	14.7 (9.0)	11.1 (9.2)	10.4 (11.6)	17.5 (7.4)	20.3 (4.0)
	Range	1-26	2-24	1-26	7-26	15-24
Time of interview	Mean (SD)	20.1 (10.7)	26.0 (12)	27.3 (14)	12.9 (3.6)	16.1 (2.2)
	Median	16.1	22	22.2	13.7	15.1
	Range	8-47	14-41	14-47	8-17	15-19
Length of hospital stay up to time of interview (weeks)	Mean (SD)	3.3 (6.0)	4.6 (7.2)	5.5 (10.3)	1.7 (0.7)	2.6 (3.5)
	Median	1.8	2.1	1.9	1.9	1.4
	Range	0-26.3	0-19.3	0-26.3	0.6-2.4	1-2.3

Table 5. Demographic characteristics of the sample

		Total Sample	Fetal Cardiac Diagnosis		Postnatal Cardiac Diagnosis	
Variable	**Mean and Range**	Parents (*N*=25)	Mothers	Fathers	Mothers	Fathers
Age (years)	Mean age (SD)	34.3 (5.1)	34.0 (3.3)	34.7 (1.6)	30.7 (4.6)	40.5 (6.8)
	Range	25-49	28-36	32-37	25-39	33-49
Variable	Level	Parents (*N*=25)	Mothers	Fathers	Mothers	Fathers
Relationship Status[#]	Together[#]	24	5	6	9	4
	Separated	1	1	0	0	0
Education	University degree	13	2	5	3	3
	No university degree	9	3	1	4	1
Gross annual household income[*]	< 104,000	14	4	4	5	2
	>104,000	7	1	1	3	2
Country of birth	Australia	17	4	5	5	3
	Other	5	1	1	2	1
Language spoken at home	English	24	6	6	8	4
	Other	0	0	0	0	0
Number of children	1	7	1	1	3	2
	2	8	2	2	3	1
	3	3	1	1	1	0
	4+	7	2	2	2	1
Planning for another child	Yes	13	3	5	3	2
	Unsure	5	0	0	0	2
	No	4	2	1	1	0

[#] Includes both married couples as well as couples in a long-term, committed relationship.

* Please note: Some participants chose not to supply all demographic information. Due to missing data not all numbers will add to the total sample size.

Table 6. Infant clinical characteristics (*N*=15)

		Fetal Diagnosis (*n*=7)		Postnatal Diagnosis (*n*=8)	
Variable	**Level**	Mean	*SD*	Mean	*SD*
Gestational age at fetal diagnosis (weeks)	Mean (SD)	22.8	(5.5)	_	_
	Median [Range]	21.1	[17-34]		
Age at postnatal diagnosis (weeks)	Mean (SD)	_	_	5.1	(4.3)
	Median [Range]			7.1	[0-10]
Age at first surgery (weeks)	Mean (SD)	3.3	(7.0)	8.7	(5.4)
	Median [Range]	0.6	[0.3-19.1]	10.1	[0.9-17.1]
Variable	**Level**	*n*	(%)	*n*	(%)
Sex	Male	3	(43)	5	(63)
	Female	4	(57)	3	(38)
Cardiac abnormality	Tetralogy of Fallot	1	(14.3)	2	(25)
	Single Right Ventricle - complex	1	(14.3)	0	0
	Single Left Ventricle	2	(28.6)	0	0
	Coarctation of Aorta	1	(14.3)	0	0
	Transposition of the Great Arteries				
	Simple	1	(14.3)	2	(25)
	Complex	1	(14.3)	0	0
	Patent ductus arteriosus	0	0	1	(12.5)
	VSD	0	0	3	(37.5)
Single ventricle	Yes	3	(42.9)	0	(0)
	No	4	(57.1)	8	(100)
Cyanotic	Yes	6	(85.7)	4	(50)
	No	1	(14.3)	4	(50)
Baby went home before first surgery	Yes	3	(42.9)	6	(75)
	No	4	(57.1)	2	(25)
Surgery in neonatal period (infant <30 days old)	Yes	6	(85.7)	2	(25)
	No	1	(14.3)	6	(75)
Number of surgeries in first year of life	1	3	(42.9)	7	(87.5)
	2	3	(42.9)	1	(12.5)
	3	1	(14.3)	0	(0)
Highest surgical complexity (RACHS)	1-2	1	(14.3)	4	(50)
	3-4	5	(71.5)	4	(50)
	5-6	1	(14.3)	0	0
	Mean (SD)	3.6	(1.3)	2.4	(0.7)
	[Range]		[2-6]		[1-3]
Cardiopulmonary bypass[#]	Yes	6	(85.7)	6	(75)
	No	1	(14.3)	2	(25)

[#]Cardiopulmonary bypass required for at least one surgery in first year of life. *Other includes: Disconnected Right Pulmonary Artery, AVSD, ALCAPA, PDA

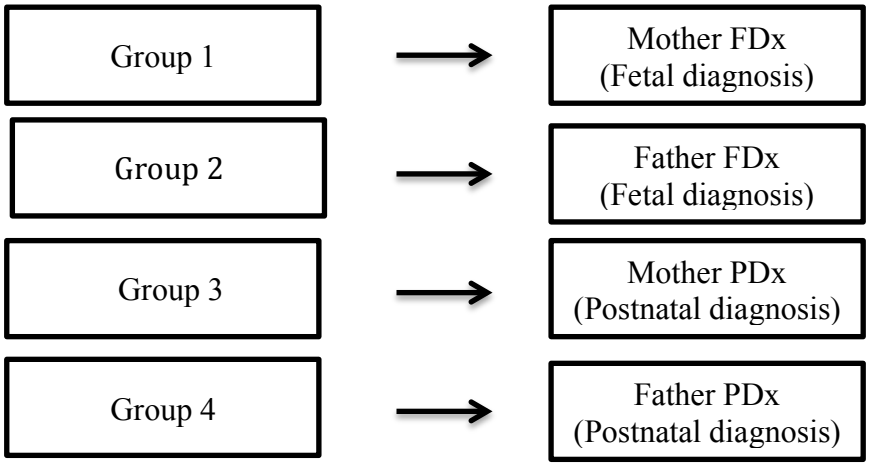
Table 7. A conceptual summary of the main components of the Lazarus and Folkman model of Stress and Coping evident

Major Components of the Lazarus and Folkman Model of Stress and Coping	Participant Quotations as Evidence
Primary Appraisal Diagnosis is significant	*Devastated, shocked and devastated. It was the worst moment.* P18
Secondary Appraisal	*'well we prepared ourselves for beyond that, not sort of how you would feel about giving birth and then that baby being taken away, that was sort of really hard to deal with. Yeah but otherwise it's being ... can be.'* P36
Problem-Focused Coping Planful Problem Solving	*'They were the more difficult things because I just wanted to tick the boxes of what I knew had to be done so we could get home'* P45
Emotion-Focused Coping Distancing	*'...I had my feet and hands out wanting to push the operation away'* P18
Seeking Emotional Support	*'My mum. Every morning she would ring me and tell me just to go about my business and enjoy my babies and just keep living life.'* P32
Escape-Avoidance	*'We didn't tell a lot of people though. I didn't tell my dad and his wife or any of my siblings...'* P32
Meaning-Focused Coping Benefit Finding	*'...you probably appreciate your child more than what you would if you hadn't have gone through that.'* P76
Benefit Reminding	*'All that - all the hard road to get to that point was worth it because we got to bring him home.'* P15-father
Adaptive Goal Processes	'*Obviously I don't want him playing rugby or something like that but he can still be an active child.*' P61
Reordering Priorities	*'Yeah I suppose that just yeah it takes over, everything else is irrelevant....'* P76
Infusing Ordinary Events with Positive Meaning	*'I like bath time, like giving her her baths, for no real reason other than I know it calms her every time.'* P82
Confrontive Coping	*'...I don't know, just this elephant kind of breakthrough where I had a lot of force, you know, and they would have had to restrain me at that point.'* P56
Accepting Responsibility	*'That I'd carried him and I'd given him, you know I hadn't made him well.'* P46-50
Self Controlling	*'... but you know you kind of hold back some of that emotion, you don't want to just fully expose how you feel sometimes.'* P36

Table 8. Differences in experiences between participants as a result of time of diagnosis

Theme	Fetal Diagnostic Group	Postnatal Diagnostic Group
Feeling prepared	Generally felt prepared, had the information they needed	Did not feel prepared, did not have the information they needed
	Felt they had access to the best medical care available	Mothers PD appraised the diagnosis as a bombshell, life changing
	Utilised the internet for information	Mothers PD did not know what ways going to happen
		Mothers PD felt helpless and out of control
Meaning-focused coping	Engaged in adaptive goal processes whereby they accepted the limitations of their child's future	Engaged in benefit finding whereby they felt their relationships with partner and family had been strengthened as a result of their experiences
Emotion-focused coping - Distancing	Engaged in distancing specifically went on as if nothing happened	Engaged in distancing specifically didn't let it get to me/ refused to think about it too much and made light of the situation/refused to get too serious
Sources of reassurance	Reliance upon faith	Reported gratitude that the stressful experience was short term, as opposed to other families in the hospital who faced lifetime challenges
		Took lots of photos of the baby
		Reported having an instant bond, connection and trust with the baby

Figure 1. Participant group categorisation.



Appendix A – Model of Stress and Coping proposed by Lazarus and Folkman (1984) and revised by Folkman (2000)

Primary Appraisal occurs when an individual is initially faced with a stressful situation (Folkman, 2010). The individual either consciously or unconsciously appraise the situation, determine if it is significant and important to them and the personal impact it is likely to have (Folkman, 2010). This appraisal is influenced by the individuals, values, priorities and ambitions (Folkman, 2010).

The individual conducts **a Secondary Appraisal** after they have appraised the situation as significant (Folkman, 2010). A secondary appraisal involves the individual evaluating their ability to cope with the situation based on their capacity to either: control, prevent, change or cope with the stressful situation (Folkman, 2010).

Problem-focused coping involves taking active, analytic and task-oriented steps to solve, address or combat the stressful situation (Folkman, 2010). Examples of problem-focused coping include planful problem solving, gathering information, confrontive coping (i.e., fighting for what you want or need), gathering necessary resources and generating plans (Folkman, 2010).

Emotion-focused coping involves the regulation of emotions that are elicited by the stressful situation (Folkman, 2010). Examples of emotion-focused coping include distancing, escape-avoidance, seeking social and emotional support and self-controlling emotions (Folkman, 2010).

Folkman has only recently added **Meaning-focused coping** to the model of stress and coping (2000). It has been included in order to recognise the role of positive emotions in a stressful situation (Folkman, 2000). It has been defined as '*appraisal-based coping in which the person draws on his or her beliefs (e.g., religious, spiritual or beliefs about justice), values (e.g., "mattering"), and existential goals (e.g., purpose in life or guiding principles) to motivate and sustain coping and well-being during a difficult time'* (Folkman, 2008, p. 7) and is made up of five different components:

1. **Benefit finding** is the most common type of meaning-focused coping reported. It involves an individual's assessment of the situation stating that it has helped them to grow in personal traits such as knowledge or ability, has helped them to recognise what it important in life and has strengthened their relationships or beliefs.

2. **Benefit reminding** involves an individual consciously reminding themselves of the benefits or positives that will likely result from the stressful situation.

3. **Adaptive Goal Processes** involves the individual developing new goals, leaving old and now unattainable goals behind and actively striving towards achieving the new goals.

4. **Reordering priorities** involves the individual making either conscious or unconscious changes in what are the important things in life. By reordering priorities the individual is able to recognise what they need to or want to be working towards and to act in a manner that can best achieve those goals.

5. **Infusing Ordinary events with positive meaning** involves an individual reporting about an ordinary event that has occurred and deliberately giving it a positive meaning. By doing this, the individual is able to generate something positive out of an event that would otherwise be ordinary and insignificant.

Appendix B – Study Package sent to all participants

(Printed on either SESIAHS or the Children's Hospital at Westmead letterhead)

PARTICIPANT INFORMATION SHEET

Title of this research study: The experiences and needs of parents who find out that their baby has a heart abnormality.

Researchers responsible for this study:

Dr Nadine Kasparian, School of Women's and Children's Health, University of NSW. Ph: 1800 814 403.
Dr Edwin Kirk, Department of Medical Genetics, Sydney Children's Hospital, Randwick. Ph: 9382 1704.
Professor Bryanne Barnett, Park House for Children and Families, Sydney West Area Health Service. Ph: 9827 8011.
A/Professor Gary Sholler, Adolph Basser Cardiac Institute, The Children's Hospital at Westmead. Ph: 9845 2345.
A/Professor David Winlaw, Adolph Basser Cardiac Institute, The Children's Hospital at Westmead. Ph: 9845 3063.

What is the purpose of this research?

You are invited to take part in a research study on the experiences and needs of parents who find out that their baby has a heart abnormality. The purpose of this study is to learn more about the experiences of mothers and fathers in this situation and thus, to identify ways in which we can improve the services offered to families affected by childhood heart disease.

Who is being invited to participate?

This research study includes mothers and fathers who have found out that their baby has a heart abnormality. The baby's heart condition may have been identified during pregnancy, or it may have been diagnosed after the baby was born. Some participants will be expectant parents who have not given birth to their baby yet, while others may have a baby who is almost one year old. All parents will be aged 18 years or over, and will have seen a paediatric cardiologist from either the Sydney Children's Hospital or the Children's Hospital at Westmead. This study is open to single parents, as well as parents who are married or in a committed relationship.

What are participants being asked to do?

If you decide to take part in this study we ask that you:

- Read and sign the Consent Form on page 3;
- Complete the yellow Participation Card;
- Return the signed Consent Form and Participation Card in the reply paid envelope provided;
- Participate in one face-to-face interview and one telephone interview with a member of our research team.
- Fill in one brief survey.

These interviews are a space for you to think and talk about your experiences since finding out about your baby's heart condition. They are designed for us to understand the thoughts and emotions you have experienced since this time about yourself, your baby, and your baby's medical care. We would also like to learn more about the types of services (if any) that you have used during this time, and whether you found these services helpful. If possible, the first interview will be held at either the Sydney Children's Hospital at Randwick or the Children's Hospital at Westmead, depending on which location is most convenient for you. We will arrange free parking for you at either of these locations and will also reimburse your travel expenses up to the value of $20; however, if you are unable to travel to the interview venue we can arrange a telephone interview for you instead.

(Printed on either SESIAHS or the Children's Hospital at Westmead letterhead)

PARTICIPANT INFORMATION SHEET (continued)

The second interview will take place two weeks later and will be conducted over the telephone. Each discussion is expected to last about 50 minutes, and will be arranged at a time that is convenient for you. You and your baby's father (or other primary caregiver) may choose to attend each interview together, or you may prefer to attend the interviews individually – this decision is completely up to you. With your permission we will tape-record these discussions so that we have an accurate record of your views and experiences. Parents will still be able to take part in this study if they choose not to have their interview audio-taped.

We will also ask you to complete one brief survey, which will take no more than 5 minutes for you to complete. This survey will ask simple questions about your background (for example, your age and marital status), as well as how you have been feeling in the past week.

What will happen to the tape-recordings?

All information collected during this study is confidential and will be stored in secure databases or locked filing cabinets that can only be accessed by the researchers working on this project. No information that you provide and that identifies you will be passed on to any other person without your explicit consent, except as required by law. Results from this study will only be presented to the scientific community and to the public in ways that protect the confidentiality and anonymity of participants. Information will be stored for a minimum of seven years from the end of the study, and then disposed of by shredding of any paper documents and erasing of the tapes.

Are there any benefits associated with this study?

There are no known benefits associated with participating in this study. We hope, however, that the results from this study will help us to improve the services offered to families affected by childhood heart disease.

Are there any risks associated with this study?

It is possible that talking about your baby's heart condition may cause you to feel upset, worried or distressed. The research team will be available to talk about any worries or concerns you may have. You can contact a member of the research team by calling the free-call study telephone number: 1800 814 403.

Do I have a choice?

Your participation in this study is completely voluntary and you can end your participation at any time. Whether or not you choose to take part in this study will not have any affect on the medical care that your infant receives now, or in the future.

What if I need more information or if I have problems with any aspect of the study?

If you have any questions about this study, or if you would like further information, please contact Dr Nadine Kasparian at the Prince of Wales Hospital on: 1800 814 403.

This study has been reviewed by the Human Research Ethics Committee (Health and Medical) of the University of Wollongong. If you have any concerns or complaints regarding the way this research has been conducted, you can contact the University of Wollongong Ethics Officer on (02) 4221 4457.

(Printed on either SESIAHS or the Children's Hospital at Westmead letterhead)

CONSENT TO PARTICIPATE IN RESEARCH

Title of research study: The experiences and needs of parents who find out that their baby has a heart abnormality.

Researchers: Nadine Kasparian, Edwin Kirk, Bryanne Barnett, Gary Sholler and David Winlaw.

I have read and understood the Participant Information Sheet and have had the opportunity to ask the researchers any questions I may have had. I understand that my participation in this research study will involve one face-to-face interview and one telephone interview with a member of the research team. These discussions are expected to last about 50 minutes each and will be audio-taped with my permission.

I understand that all information collected during this study is confidential and will be stored in secure databases or locked filing cabinets that can only be accessed by the researchers working on this project. I also understand that I am free to withdraw from this study at any time, and that this decision will not have any affect on my baby's medical care now or in the future.

NAME OF PARTICIPANT: _____
(Please print)

SIGNATURE OF PARTICIPANT: _____ Date: _____

A copy of this consent form will be given to you to keep.

Appendix C – Interview Discussion Guide

Discussion Guide

Introduction

• Thank parent for agreeing to take part in this study.

• This interview is a space for you to think and talk about your experiences since finding out about your baby's heart abnormality. My role is to listen, and to ask questions to help us understand the thoughts and feelings you have experienced since your baby's diagnosis.

• At times, you may find some of the questions challenging, but the reason I ask these questions is so that we can learn as much as we can to improve the services offered to families affected by childhood heart disease.

• Please feel free to use this discussion to talk about any aspect of your experience.

• With your permission, our discussion will be audio-taped so that we can keep a record of what is said, and this will be kept securely and treated as confidential.

• Clarify that the interview will take about 50 minutes and ask whether the parent has any questions.

Discussion Guide

To start, acknowledge where the parent is at in terms of their baby's heart condition. For example, "I know that you have only just found out that your baby has a heart abnormality". Or, "Your baby was born 3 weeks ago". Allow the parent time to talk about this.

As appropriate, ask about the following, allowing parents as much time as they need to talk about their experiences:

During pregnancy

• How did you come to know that you were having a baby?

• Had you planned to fall pregnant or was it a surprise?

• Was this your first pregnancy? (Any miscarriages before this pregnancy?)

• How did you feel physically during your pregnancy? Did you feel unwell at any stage?

• When did the pregnancy seem real to you?

• What were your impressions about the baby during pregnancy?

Diagnosis

• How did you come to know that there was a problem with your baby's heart?

• Can you describe what this time was like for you?

Explore experiences of sadness, grief, confusion, anger, anticipation, numbness, interrupted sleep.

• Did anyone else attend the medical appointment with you? What was it like to have/not have someone with you?

• What three words would you use to describe your experiences at this appointment (diagnosis)?

• Was there something that you found particularly helpful in getting you through this time?

• How do you think your partner coped?

Thinking about your unborn baby

• Did you know that heart defects were possible with babies?

• Did you know that some heart problems could be detected during pregnancy?

• While you were pregnant, were you able to think about your baby's heart?

• How did you imagine this condition might affect your baby, if at all?

• Were you about to share these thoughts and feelings with your partner? Why or why not?

• When did you first tell your family or friends about your baby's heart condition? How did they respond?

• How have your thoughts or feelings changed over time since first learning of your baby's heart condition?

What might have caused your baby's heart abnormality

• Parents sometimes wonder or have ideas about why they have a baby with a heart condition. Do you ever wonder about anything like this?

• Do you ever wonder about what might have caused your baby's heart abnormality?

Birth

• Can you tell me about your [or your partner's] labour and delivery?

• What did you think or feel when you first saw your baby?

• How did your partner feel?
• Did you or your baby have any problems in the first few days after birth?
• What was this time like for you and your baby?

Experiences of hospital, surgery and/or treatment

• Can you tell me about your experience of your baby's time in hospital? (Also ask about surgery)
• What was this time like for you and your baby?
• Were you able to share your thoughts and feelings at this time with your partner?
• How did you feel about the care that you and your baby have received from the hospital staff?
• How do you imagine your other children might have experienced this time?
• How were other aspects of your life affected?
E.g., physical health, mental health, sleep, work, finances, friendships, family relationships.
• Does your baby need to have any more surgeries in the future? How do you feel about this?
• What do you imagine his or her health will be like in the future?
• How did you mother her in the hospital

First few weeks at home

• How would you describe the first few weeks at home with your baby?

Reflecting on your experiences

• Of all the things that you and your baby have been through, what has been the most difficult experience for you?
• Was there something that you found helpful in getting you and your baby through this time?
• Out of all the people that you've come across, who do you think has been the most helpful? Why?
• Who do you think has been the least helpful? Explore how this could have been improved.
• Have you had the opportunity to make contact with other parents who have had similar experiences? How? Do you think this has been [or was/could be] helpful?

If things had been different

• It is possible that some people may think about what life may have been like if they hadn't fallen pregnant or if things had been different. Have you ever felt this way?

Your baby as a little person

• Can you tell me a little bit about your baby?
• What is it like to be a parent to this baby?
• Does your baby seem to have a regular routine?
• How would you describe his or her personality?
• Does he or she get upset often?
• Do parents talk about any major milestones in their baby's development?
• How were these milestones achieved? With difficulty?
• How do parents perceive their baby in relation to other babies?
• Can you tell me about a favourite experience that you and your baby have shared?

When coming to the end of the interview, let the parent know that "in the next 10 minutes we are coming to the end of today's discussion. Is there anything that you would like us to talk about before we end?"

Your experience of this interview

• How has it felt for you to be a part of this interview today?
• Was it what you have expected or imagined?

Closing

• Thank the participant for sharing their thoughts and experiences.
• Did our discussion raise any questions for you? Would you like to arrange a time to talk with a member of the research team about anything?
• Explain questionnaire will be sent in mail in next few days, and I will call again when I have received their completed questionnaire.
• Give participant Study Contact Card.