A DESCRIPTIVE, CROSS-SECTIONAL, CORRELATIONAL EXPLORATION OF PERCEIVED STRESS, QUALITY OF LIFE, AND FAMILY FUNCTIONING IN PARENTS OF A CHILD WITH CONGENITAL HEART DISEASE: THE PINCHED STUDY

A DISSERTATION IN
Nursing

Presented to the Faculty of the University
of Missouri-Kansas City in partial fulfillment of
the requirements for the degree

DOCTOR OF PHILOSOPHY

by

Mary Gregory

B.S.N., Missouri Western State University, 2003
M.S.N., University of Missouri-Kansas City, 2009

Kansas City, Missouri
2019

A DESCRIPTIVE, CROSS-SECTIONAL, CORRELATIONAL EXPLORATION OF PERCEIVED STRESS, QUALITY OF LIFE, AND FAMILY FUNCTIONING IN PARENTS OF A CHILD WITH CONGENITAL HEART DISEASE: THE PINCHED STUDY

Mary Gregory, Candidate for the Doctor of Philosophy Degree

University of Missouri-Kansas City, 2019

ABSTRACT

Background: As survival rates for infants born with severe forms of cardiac disease improve, attention is directed to evaluating factors that affect the child's short- and long-term outcomes, including parental stress, quality of life, and family functioning. Facing the unique struggles of having a child with congenital heart disease (CHD) can often result in high stress for the child, their parents, and other family members and may result in adverse effects in family functioning. Mothers of a child with CHD tend to report higher levels of stress and poorer quality of life and family functioning when compared to mothers of heart-healthy children or children with other chronic illnesses. Paternal perspectives when having a child with CHD have been understudied in comparison to mothers of a child with CHD.

Purpose: The purpose of this descriptive, correlational, cross-sectional study was to explore parental perceptions of stress, quality of life, and family functioning when having a child with CHD. Factors that influence parental stress, quality of life, and family functioning

when having a child with CHD are described, and relationships between the perceived factors and comparisons between the perceptions of mothers and fathers were made. relationships being examined. McCubbin and Patterson's (1983a, 1983b) double ABCX theory of family adjustment and adaptation was chosen as the framework for this study.

Methods: A purposive sample of 62 parents of a child with a CHD below six years of age, who had received neurodevelopmental care from the Cardiac Neurodevelopmental program at Children's Mercy Hospital in Kansas City, Missouri were included in the study. The parents completed the following instruments: The Pediatric Inventory for Parents (PIP), The Pediatric Quality of Life Inventory™ Family Impact Module (PedsQL™ FIM), and a demographics survey.

Results: Thirty-one parent pairs participated in this study. The mean age for mothers and fathers were 36.68, ±5.353 and 38.48, ±5.941, respectively. Race and ethnicity of the parent population was largely homogeneous, with mothers (n = 31, 90.3%) and fathers (n = 31, 93.5%) being of White race, and, of parents who reported ethnicity, mothers, (n = 19, 100% White) and fathers, (n = 21, 54.8% White; 12.9% Hispanic or Latino). The mean education level for mothers and fathers was just under that of a bachelor's degree (mothers μ = 9.90, SD= 1.720) (fathers μ = 9.61, SD = 1.706). Parent pairs had a mean relationship length of 11.37 years. The children with CHD of the participating parents were mostly male (n = 19, 61%). The child's mean age at time of parent survey completion was 4.83 years and have 18 different fundamental CHD diagnoses among the sample.

Among 62 parents of 31 children with CHD, all subscale and summary scale median stress scores for fathers fell within the low stress range except for total frequency of stress (n =31, Mdn = 86.00, IQR = 35), which fell within the moderate stress range. Mothers reported

median scores in the low stress range for all subscales except for the following subscales that had scores in the moderate stress range: emotional distress frequency (n = 31, Mdn = 33.00, IQR = 118), emotional distress difficulty (n =31, Mdn = 37.00, IQR = 20), total frequency (n = 31, Mdn = 86.00, IQR = 47), and total difficulty (n = 31, Mdn = 86.00, IQR = 47). There was a statistically significant difference (Z = -2.30, p = 0.02) in the role functioning subscale where fathers (n = 31, Mdn = 16.00, IQR = 10) reported less difficulty in role functioning than mothers (n = 31, Mdn = 21.00, IQR = 16). In regards to quality of life, fathers reported high levels of quality of life in all subscales and summary scales, and mothers reported high levels of quality of life in all subscales except emotional functioning, worry, and the health related-quality of life (HRQOL) summary scale, which were all in the moderate range. Statistically significant scores were found in emotional functioning, where fathers reported statistically significant (Z = -2.52, p = 0.01) better emotional functioning (n = 29, Mdn = 450.00, IQR = 162) in comparison to mothers (n = 29, Mdn = 350.00, IQR = 250), and in communication, where fathers reported statistically significant (Z = -2.38, p = 0.02) better communication (n = 29, Mdn = 275.00, IQR = 100) in comparison to mothers (n = 29, Mdn = 225.00, IQR = 137.5). There were no statistically significant differences between family functioning scores between mothers and fathers. All measured factors of parental stress and quality of life were found to have statistically significant relationships with family functioning ($p \leq 0.05$). Regarding the relationship between stress and family functioning, fathers of a child with CHD reported lower mean scores in every PIP subscale and summary scale compared to mothers. Regarding the relationship between quality of life and family functioning, the PedsQL-FIM quality of life summary score and family functioning summary scores are positively correlated ($r(58) = 0.84$, p = 0.00).

There were no statistically significant relationships between the severity of the child's heart defect type, as measured by the STAT and parent reports of stress, quality of life, and family functioning ($p < 0.05$). Additional analyses determined time since most recent cardiopulmonary bypass surgery had a positive association on communication of both parents ($r(58) = 0.275$, $p = 0.03$), and fathers of a child with CHD ($r(28) = 0.396$, $p = 0.03$). Regarding the frequency of past cardiopulmonary bypass (CPB) procedures, the only statistically significant difference ($U = 3.00$, $p = 0.007$, $r = -0.63$) found was among fathers of a child who had had one CBP procedure and fathers of a child who had had four ($n = 4$, $Mdn = 25.00$, $IQR = 4$) CBP surgeries when compared to those whose child had had only one CBP surgery ($n = 14$, $Mdn = 17.00$, $IQR = 7$). Having a child with CHD and abnormal brain imaging negatively influenced many aspects of their parents' quality of life and family functioning ($p < 0.05$): physical function ($r(58) = -0.296$, $p = 0.02$); social functioning ($r(58) = -0.254$, $p = 0.05$); worry ($r(58) = -0.281$, $p = 0.03$); daily activities ($r(58) = -0.314$, $p = 0.01$); parent HRQL summary score ($r(58) = -0.260$, $p = 0.04$); family functioning summary score ($r(58) = -0.260$, $p = 0.05$); and total FIM score ($r(58) = -0.267$, $p = 0.04$).

When their child had received early intervention services, there were statistically significant associations in all of the parents' reports of worry, ($r(58) = -0.281$, $p = 0.03$); and daily activities, ($r(58) = -0.328$, $p = 0.01$). For gender-based sub groups, fathers had statistically significant associations with worry ($r(29) = -0.374$, $p = 0.04$), and mothers had statistically significant associations found with the daily activities subscale ($r(29) = -0.393$, $p = 0.03$).

Discussion: The differences between fathers and mothers of a child with CHD were not clinically meaningful. The descriptive statistics for scaled and summary scores indicated

that parents who report better outcomes in their stress and QOL also report better overall family functioning and vice versa. These results indicated that parent perceptions of their stress, QOL, or family's functioning were not significantly impacted by the severity of the child's heart defect; therefore, severity of CHD type should not be used to predict which parents may experience high levels of stress or poorer QOL and family functioning. Results also demonstrated as more time passed, QOL for parents improved and may serve as an indicator of parents developing bonadaptation related to their child's health condition. Having a child with CHD and a known developmental delay or brain injury may serve as a better indicator for identification of parents and families who will benefit from supportive interventions.

Conclusions: This pilot study demonstrated feasibility for additional research about the experiences among parents of a child with CHD to understand their needs for support, and to determine if fathers report similar outcomes as mothers, who are much more prevalent in research addressing parental outcomes when having a child with CHD. Longitudinal and interventional studies will assist in determining timing and effectiveness of supportive interventions for parents of a child with CHD. Parent-supportive policies will benefit from additional father-inclusive research and advocacy.

Keywords: parents, stress, quality of life, family functioning, congenital heart defect, congenital heart disease, gender differences

APPROVAL PAGE

The faculty listed below, appointed by the Dean of the School of Nursing & Health Studies, have examined a dissertation titled, "A Descriptive, Cross-Sectional, Correlational Exploration of Perceived Stress, Quality of Life, and Family Functioning in Parents of a Child with Congenital Heart Disease: The PinCHeD Study," presented by Mary R. Gregory, candidate for the Doctor of Philosophy degree, and hereby certify that in their opinion it is worthy of acceptance.

Supervisory Committee

Sue Lasiter, Ph.D. RN, Committee Chair
UMKC School of Nursing and Health Studies

Elizabeth Willen, Ph.D.
Children's Mercy, Kansas City
UMKC School of Medicine

Cynthia L. Russell, Ph.D. RN
UMKC School of Nursing and Health Studies

Katherine Mussatto, Ph.D. RN
Associate Professor
Milwaukee School of Engineering University School of Nursing

An-Lin Cheng, Ph.D.
UMKC School of Medicine

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LIST OF ABBREVIATIONS

ASD	Atrial septal defect
CHD	Congenital heart defect/disease
CND	Cardiac neurodevelopment/neurodevelopmental
DILV	Double inlet left ventricle
DORV	Double outlet right ventricle
EI	Early intervention
FCCHD	Fathers of a child with congenital heart defect/disease
FMLA	Family Medical Leave Act
HLHS	Hypoplastic left heart syndrome
HRQOL	Health related quality of life
IVS	Intact ventricular septum
IQR	Interquartile Range
MCCHD	Mothers of a child with congenital heart defect/disease
ND	Neurodevelopmental difference/delay
PA	Pulmonary atresia
PCCHD	Parents of a child with congenital heart defect/disease
PCOD	Parents of a child with other disease
PedsQL-FIM	Pediatric Quality of Life-Family Impact Module
PHC	Parents of a healthy child
PI	Primary investigator
PinCHeD	Parents of a child with Congenital Heart Disease
PIP	Pediatric Inventory for Parents

PPE	Postage paid envelope
PPND	Paternal perinatal depression
PTSD	Post-traumatic stress disorder
QOL	Quality of life
TAPVC	Total anomalous pulmonary venous connection
TGA	Transposition of the great arteries
TOF	Tetralogy of Fallot

ACKNOWLEDGMENTS

This research was supported by a grant from the University of Missouri- Kansas City's Women's Council, Shannon Stone Award and Presidents and Past Presidents General Assembly of Greater Kansas City Award II.

APPRECIATION PAGE

First, I need to thank God for giving me the strength, endurance, and wisdom during this period of my life. I would not have been able to have done this without his loving guidance. I would like to thank the parents who participated in this research. I would like to thank the leadership, providers, and patients of the Developmental and Behavioral Department, and Ward Family Heart Center, specifically the Cardiac Neurodevelopmental Program at Children's Mercy Hospital for supporting me during this endeavor. There are no words to encompass my gratitude.

During this lengthy educational, personal, and professional journey, I have been supported and mentored by so many individuals I feel a list here may inadvertently omit someone; therefore my words are for each of you. Thank you for encouraging me, pushing me further than I thought I could go, motivating and mentoring me. My special, heart-filled thanks are extended to my children, Adam, Katie, and Gavin. You have endured the brunt of my stress-filled days, homework-filled nights, and other sacrifices in my inability to balance everything as well as I wish I could have. My friends, specifically, Ronda, Crystal, Holly, Tammy, Rachel, and so many more, you have no idea how much I value and cherish you all and your efforts to make sure I made time for a break from work and homework. You ladies are also my family. To my family in Ohio and Michigan, thank you for believing in, loving, and supporting me across the miles. To my father, thank you for exemplifying the truest form of fatherhood. Lastly, to my mother, who passed away as I was editing the final chapters of this work, thank you for just being my mother. I know you are always with me. I love and appreciate you all.

My educators, mentors, and committee members (in no particular order), Dr. Sue Lasiter, Dr. Ahn-Lin Cheng, Dr. Christy Roberts, Dr. Patricia Kelly, Dr. Cynthia Russell, Dr. Kathleen Mussatto, Dr. Elizabeth Willen, and Dr. Jennifer Hunter have tended me as they would have tended a well-cared-for garden. They have found in me a weedy lot but saw potential! They helped me develop soil for a fine foundation, ensuring the correct seeds were planted, and assessed my progress and growth. They allowed autonomy and provided support while challenging me. This dissertation is the cherished bloom of my hard work and their guidance. Thank you all for your wisdom, time, faith, and mentorship.

CHAPTER 1

INTRODUCTION

Chapter 1 provides the context for this study of parents with a child who has congenital heart disease (CHD). The prevalence and pathophysiology of CHD, descriptions of neurological sequelae, and types of neurodevelopmental sequelae frequently experienced by children with CHD are explained. Attention is given to potential challenges parents of a child with CHD face, and impact on parental stress, quality of life, and family functioning are discussed. This study addresses the under-representation of fathers in the CHD literature by sampling parent dyads and comparing gender-based results. Therefore, the importance of including fathers of children with CHD in research, the psychological and emotional well-being of fathers, and the socioeconomic factors that lead to increasing father presence in child-rearing are addressed. Finally, the significance, innovation, study purpose, research questions with hypotheses are provided.

Congenital Heart Disease

Congenital heart disease is one of the most common birth defects, affecting approximately 40,000 neonates each year; and, of those infants born with CHD, 25% of the defects are considered critical in nature with accompanying high mortality rates (Centers for Disease Control and Prevention, 2016; Oster et al., 2013). Congenital heart disease consists of an array of anomalies involving malformations of the heart and related vessels and their functions that develop in utero and are diagnosed prenatally or even as late as adulthood (American Heart Association, 2019; Ottaviani & Buja, 2016). These malformations in cardiac vascularization and cardiac function adversely affect fetal and neonatal brain development, which may lead to brain injury and/or neurodevelopmental delays (Claessens,

Kelly, Counsell, & Benders, 2017; Ortinau et al., 2012). Advances in surgical techniques and medical management have lowered mortality rates for even the most complex CHD types (Mahle, 2011; Mahle et al., 2013; Marino et al., 2012; Pasquali et al., 2012) resulting in an increased number of infants and children returning home to be cared for by their families.

Neurological Sequelae of Congenital Heart Disease

There is no research to date that has identified the exact risk of neurological injury in infants with CHD. However, there are several pathophysiological conditions that contribute to the risk of neurological injury. Chen et al. (2009) reported an increase in periventricular leukomalacia, a form of brain injury often diagnosed in children with CHD, from 16% pre-cardiac surgery to 48% after cardiac surgery. Inadequate blood oxygenation or impaired cerebral blood flow in utero or after delivery has been shown to negatively impact brain development (Kaltman, Di, Tian, & Rychik, 2005; Licht et al., 2004). Cardiac disease and complications during medical management is a leading cause of stroke in children and may be left undetected because it can be clinically silent during infancy (Chen et al., 2009; Sinclair et al., 2015). For children with CHD, the prevalence and severity of developmental delay increases with the complexity of the heart defect. Additionally, whether acquired as a fetus or post-delivery, the degree of leukomalacia, hypoxic injury, and stroke coupled with the presence of co-morbid conditions such as genetic syndromes, premature birth, drug exposure, and lengthy hospital stays (Donofrio, Duplessis, & Limperopoulos, 2011; Limperopoulos et al., 1999; Limperopoulos et al., 2000; Mahle & Wernovsky, 2001; Marino et al., 2012) further increase the risk of neurological defecits.

Neurodevelopmental Sequalae of Congenital Heart Disease

Neurodevelopmental delays (ND) are a critical issue for children with CHD. Research on neurodevelopmental outcomes indicate nearly half of children who required cardiac interventions such as cardiac catheterizations or surgery to repair or palliate their defect have exhibited neurodevelopmental delays (Verrall et al., 2019). There is a wide spectrum in the type, duration, and severity of the neurodevelopmental differences found among children affected by CHD that include gross and fine motor delay, language developmental delays, cognitive impairments, social difficulties, and challenges with executive functions, attention, hyperactivity, and maladaptive behaviors (Bjarnason-Wehrens et al., 2007; Gaynor et al., 2015; Marino et al., 2012; Nathan et al., 2014; Newburger et al., 2012; Ravishankar et al., 2013; Tabbutt, Gaynor, & Newburger, 2012). Compared with the estimated prevalence for their general age-mate population, children with CHD have higher rates of language impairment (Miatton, De Wolf, Francois, Thiery, & Vingerhoets, 2007; Uzark, Spicer, & Beebe, 2009), decreased social competence (Bellinger, 2008), attention dysfunction (Hövels-Gürich et al., 2007; Shillingford et al., 2008; Shillingford & Wernovsky, 2004), and autism spectrum disorders (Antshel et al., 2007; Hultman, Sparen, & Cnattingius, 2002; Wier, Yoshida, Odouli, Grether, & Croen, 2006).

Parental Outcomes of Caring for a Child with CHD

Parental Stress

Lazarus (2006) described stress as a dynamic process that happens when an individual perceives the demands of a situation exceeds their available resources. Baum (1990) stated that stress is an uncomfortable emotional experience that can have associated physical, psychological, and behavioral changes. Whereas chronic or extreme stress can

adversely affect an individual's health, not all stress is detrimental, and some stress may produce motivation and energy to accomplish tasks (American Psychological Association, 2019b). Due to the intensity of a child's medical needs, parents often find themselves taking on caregiver roles for which they have little or no preparation (Rempel, Ravindran, Rogers, & Magill-Evans, 2012). Parents become so focused on the management and surveillance of the medical health of their child with CHD that they overlook the needs of themselves and other family members (Bishop et al., 2019; Drotar, 1997). Facing the unique struggles of having a child with CHD can often result in high stress for both the child and the parents and may result in adverse effects in family functioning (Minor, Carlson, Mackenzie, Zernicke, & Jones, 2006). Furthermore, parental stress, or aspects of it such as increased maternal worry, correlates with or predicts adverse psychosocial or behavioral outcomes in their young child with CHD (Majnemer et al., 2006; McCusker et al., 2007).

Quality of Life

According to the World Health Organization, QOL is "the individual's perception of his/her position in life in the context of culture and value systems in which he/she lives and in relation to his/her goals, expectations, standards, and concerns" (1995, p. 1). An operational definition of HRQOL is "the combined objective measure and subjective perception of an individual's physical, mental, and social functioning as he/she contributes to or is influenced by his/her current and future health status" (Paltzer, Barker, & Witt, 2013, p. 1178). Health-related QOL (HRQOL) evolved from the QOL concept due to a growing consensus that QOL was too broad and did not capture the nuanced influences of health on QOL (Haas, 1999). Parents of a child with CHD report lower QOL than parents of healthy children (Goldbeck & Melches, 2005; Lawoko & Soares, 2003a). Major adverse

consequences to parental QOL have been associated with the increased complexity of CHD type (Kahr, Radke, Orwat, Baumgartner, & Diller, 2015). For concept identification in the scope of this work, "QOL" and "HRQOL" are referred to as "QOL."

Family Functioning

Family functioning refers to the comprehensive properties of the family environment including relationships among family members and the levels of conflict, cohesion, adaption, communication quality, and organization (Alderfer et al., 2008; Lewandowski, Palermo, Stinson, Handley, & Chambers, 2010). In pediatric chronic health conditions, the impact of disease and treatment on family functioning is of significant concern given the essential role of parents and the family in assisting with the child's adaptive functioning and development (Thomasgard & Metz, 1999; Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004; Varni & Wallander, 1988). Research has demonstrated that parents of children with severe types of CHD meet criteria for psychological distress, and family functioning is negatively impacted after their child is discharged from the hospital following cardiac surgery (Helfricht, Latal, Fischer, Tomaske, & Landolt, 2008). Presence of family strain may serve as a predictor of the child's school adjustment more than the child's actual physical limitations due to their medical condition (Casey, Sykes, Craig, Power, & Mulholland, 1996; Mussatto, 2006). The systematic review provided in Chapter 2 explains that the majority of research on parental perspectives when having a child with CHD has been completed outside of the United States with mothers being more represented in the samples than fathers. The bulk of the research on parental perspectives research in nursing and medicine about relationships between parenting behaviors and the outcomes of their child with CHD has consistent recommendations for personal and family psychosocial support (American

Nurses Association, 2015; Brosig, Mussatto, Kuhn, & Tweddell, 2007; Human, 2009; McCusker et al., 2007; Vrijmoet-Wiersma, Ottenkamp, van Roozendaal, Grootenhuis, & Koopman, 2009; Wernovsky, 2008). Interventional studies have not been performed among parents of a child with CHD as researchers continue to understand the areas of family functioning most impacted and the support needs these parents experience over the life course of their child with CHD.

Fathers of Children with CHD in Psychological and Emotional Well-Being Research

Although unequally represented compared to mothers in the literature about parents of a child with CHD, fathers have reported high levels of stress, and their QOL was adversely affected when compared to fathers of healthy children or children with other types of chronic illnesses (Azhar, AlShammasi, & Higgi, 2016). Fathers of children with CHD have reported a decrease in QOL because of the time-consuming and difficult role of being the emotional support provider for their spouse or partner (Svavarsdottir & McCubbin, 1996). Studies conducted in the first three months of the child's life or prior to any cardiac repair or palliation demonstrated significantly more social functioning complaints, higher stress levels, depression, and differences in perceived mental and physical health in fathers of a child with CHD compared to fathers of healthy children (Bevilacqua et al., 2013; Utens et al., 2000). Bright et al. (2013) conducted a mixed methods, cross-sectional study that examined the relationships 63 fathers had with their infant with CHD and determined over one-third of these fathers felt closer to their infant with CHD and responded to this child's needs more quickly than their other children because of the cardiac condition and experiences the infant had already gone through. Da Silva et al. (2016) conducted a descriptive, qualitative study of 10 fathers to understand the difficulties they experienced in

day-to-day care of their child with chronic illness and found these fathers endorsed difficulties balancing child care with work, increased emotional burdens, difficulty with medication administration, hospitalizations, and sharing care with the child's mother. Werner et al. (2014) conducted a prospective, cohort study of 104 Swiss families of children with CHD to examine the influence of CHD and psychosocial factors on the family and found no differences among mother (n = 81) and father (n = 66) reports of family functioning. Werner et al. (2014) also found the presence of a genetic disorder in their child and lower levels of perceived social support among parents were associated with a greater impact on their family. In addition, Bevilacqua et al. (2013) conducted a pilot, cross-sectional correlational study on 38 Italian parental couples of infants with CHD shortly after their infant's discharge from the hospital and determined mothers experienced higher stress and depression levels than fathers and timing of diagnosis (prenatal vs. postnatal) influenced stress and depression levels similarly among parents. Utens et al. (2000) conducted a cross-sectional study on German parents (mothers = 94; fathers = 92) of a child with CHD to assess the levels of psychological distress and coping styles reported prior to elective cardiac surgery vs. elective interventional cardiac catheterization and found parents endorsed elevated levels of psychological distress and less adequate styles of coping, with mothers of a child with CHD reporting greater problems with coping and distress compared to fathers. These European studies support the need to better understand the differences among parent genders and the need for studies on American parents of children with CHD for their unique experiences

Socioeconomic Shifts Increasing Father Presence

The systematic review presented in Chapter 2 demonstrates a lack of research related to perspectives from parents of a child with CHD resulting from a sampling gap that favors mothers who tend to escort their child to doctors' visits where parental data are often gathered. Menahem, Poulakis, and Prior (2007) observed that "fathers did not seem too keen to be involved" (p. 608) in their study and speculated that fathers may have accepted their role to be their partner's support and the information they could provide was not as important or valid as the mother's. A systematic review by Sarkadi, Kristiansson, Oberklaid, and Bremberg (2008) provided evidence of the positive influences father engagement has on their child's developmental outcomes. Sarkadi et al. (2008) indicate that there has been an increase in fathers who are more involved with their child's rearing due to socioeconomic forces that negatively impact paternal employment. Paternal unemployment has led to more opportunities for fathers to stay at home and participate in the child-rearing while working mothers sustain the family income. In Petroski and Edley's (2006) conceptual exploration of the stay-at-home father, they discussed how mothers have increased their presence in the workforce, resulting in restructuring and reorganizing of the traditional family, which allowed fathers to take on increased parenting roles and functions. This is consistent with reports indicating that fathers in other developed countries spend as much, or more, time with their children than mothers (Clutton-Brock, 1991; Eibl-Eibefeldt, 1989; Whiting & Whiting, 1975). The transition of economic influence between parent genders and cultural shifts in social media, television, and internet usage by fathers for support and education were indicators that fathers have been more involved with their child's care than ever before (Raeburn, 2014; United States Census Bureau, 2016). Numerous studies have demonstrated that the change in paternal perceptions related to non-traditional attitudes to earning and

childcare are associated with more satisfying adult sexual partnerships, higher self-esteem, and greater life-satisfaction when engaged with their child (Flouri, 2005; Pleck & Masciadrelli, 2004; Sarkadi et al., 2008). In a clinical report for the American Academy of Pediatrics addressing fathers' role in the care and development of their children, fathers of children with special health care needs were found to be highly involved in child-rearing and frequently advocated for their child's medical needs even if it meant "positioning themselves in the health care system as an 'unpopular' family member" (Yogman & Garfield, 2016, p. e5).

Although fathers have been studied less frequently than mothers, there is general agreement that father involvement is equally as important as mother involvement for the child's overall development and well-being regardless of the child's health conditions (Flouri, 2005; Jackson, Frydenberg, Liang, Higgins, & Murphy, 2015). Sarkadi et al. (2008) completed a literature review of 24 articles describing the positive effects of father involvement on their child's social, behavioral, and psychological outcomes developmental outcomes. Bruce, Lindh, and Sundin (2016) performed narrative interviews with five Swedish fathers of a child with CHD to understand their lived experiences and feelings of being supported. This research determined these fathers desired the support needed to improve and increase the interactions they had with their child and to participate in child-raising (Bruce et al., 2016). Pleck and Masciadrelli's (2004), Flouri's (2004), and Sarkadi et al.'s (2008) research explain high father involvement benefited children by improving their peer relationships, decreasing problem behaviors, lowering substance abuse and criminality, predicting higher occupational mobility relative to their parents, and increasing the child's capacity for empathy. Fathers of children with CHD make vital contributions to their

families' well-being and their child's rearing and development beyond escorting them to doctor visits where data are frequently collected. The previous body of literature provides a foundation to support the need for equal representation of fathers in the research conducted on parents of a child with CHD to better understand their unique experiences and needs for support.

Areas of Opportunity

In summary, research has shown parents experience increased stress, impacted QOL, and differences in their family's functioning when having a child with CHD. We know that fathers play a key role in the development of their child and desire to be supported in a manner that allows them to participate in the raising of their children. What is not well understood or documented in the literature are the very specific aspects of parental stress, QOL, and family functioning that cause the most concern or difficulty for parents of a child with CHD. Most literature that includes father perspectives when having a child with CHD is conducted in countries other than the United States with inconsistent use of a theoretical framework to guide the studies. We also do not have a strong grasp on the differences these experiences are perceived by mothers and fathers due to the unequal representation of fathers in the CHD literature. Guided by McCubbin and Patterson's (1983a, 1983b) double ABCX theory of family adaptation and adjustment, this study used instruments that allowed participating parental pairs of children with CHD to describe specifics about the stressors they experienced and the manners and intensity their QOL and family's functioning were most impacted. The ability to identify specific aspects of perceived stress, QOL, and family functioning that are most affected in these parents may assist clinicians in determining and providing the most effective and appropriate interventions to lessen the impact a stressor has

on the parent's QOL, their family's functioning, and promote bonadaptation in these parents and families.

Significance

Although CHD is one of the most common birth defects (Centers for Disease Control and Prevention, 2016), relationships between the child's cardiac condition, the child's medical and neurodevelopmental outcomes relative to their parents' stress, quality of life (QOL), and family functioning are understudied. Additionally, studies that examine the exchange of influences that occur between health outcomes of the child with CHD and the perceptions and experiences of their parents are lacking (Brown, Wernovsky, Mussatto, & Berger, 2005; Massaro, El-Dib, Glass, & Aly, 2008; Rempel & Harrison, 2007). Medical treatment concerns for the child with CHD are still evident in the literature; however, there is an emerging view that family factors have a greater impact on the child's long-term outcomes than the heart defect or treatment of the CHD (McCusker et al., 2007). Having a child with CHD influences physical, social, emotional, and cognitive functioning, communication, worry, daily activities, and family relationship factors of parental QOL and family functioning (Hoehn et al., 2004; Jack, 2004) in ways we continue to try to understand. Chapter 2 provides a systematic review of the literature (Gregory, Prouhet, Russell, & Pfannenstiel, 2018) that examines the literature on parental outcomes among parents of a child with CHD critically and advances three gaps in the literature were in the PinCHeD study.

The first gap recognized in current research is the unequal representation among mothers and fathers in sampling that favors maternal experiences in the CHD literature (Gregory et al., 2018). Exploratory and correlational research is needed to improve our

knowledge of parental perceptions when having a child with CHD since mothers and fathers may perceive their situations differently and could benefit from different types of supports and interventions The PinCHeD study sought to address the underrepresentation of fathers in research sampling which is responsible for the knowledge gap of father perspectives in the CHD literature. When corrected, equal representation would allow for comparison of fathers' perspectives to those of mothers while eliminating as many compounding factors as possible through purposeful sampling of parent pairs from the same households.

The second gap in the literature the PinCHeD study addressed is the inconsistent use of a theoretical framework among research related to parental outcomes when having a child with CHD. Gregory et al,'s (2018) systematic review noted the use of a theoretical framework can guide the researcher in organization of the study, identification of constructs to measure, the selection of instruments, and statistical analyses to perform to understand the relationships being examined (Polit & Beck, 2008). The PinCHeD study addressed this gap through its use of McCubbin and Patterson's (1983a, 1983b) double ABCX theory of family adaptation and adjustment as a framework.

The systematic review (Gregory et al., 2018) provided in chapter 2 describes the third gap in the literature by its outlining of the extensive list of instruments used to examine parental perspectives when having a child with CHD. The use of different instruments to measure similar parental outcomes makes it difficult for comparisons among the findings. The PinCHeD study addressed this third gap in the research by aligning with the recent works of Caris et al. (2016), Kaugars, Shields, and Brosig (2018), and Bishop et al. (2019), with the administration of the Pediatric Inventory for Parents (PIP) (Streisand, Braniecki, Tercyak, & Kazak, 2001) to assess parental stress when having a child with CHD. The PIP

has been effectively used to measure parental stress when having a healthy child or a child with other chronic health conditions including diabetes (Hilliard, Monaghan, Cogen, & Streisand, 2011) and cancer (Vrijmoet-Wiersma et al., 2009). Comparison of the PinCHeD study with the Caris et al. (2016), Kaugars, Shields, and Brosig (2018), and Bishop et al. (2019) studies is more fully discussed in Chapter 5.

Innovation

This study contributed to the body of knowledge by increasing the diversity of the population of interest by assessing parents of a child with CHD from the Central-Midwest region of the United States (Kansas and eastern Missouri). Of studies on parental outcomes when having a child with CHD, most American samples are completed from a limited number of pediatric hospitals in limited geographical regions within the United States. Parents are represented from regional areas that include Northern-Central United States (Brosig, Whitstone et al., 2007; Hancock et al., 2016), Eastern United States (Bishop et al., 2019; Blume et al., 2014), and Western United States (Balkin et al., 2015; Sklansky et al., 2002). Parents of children with CHD from the central-Midwestern region of the United States, such as those included in the PinCHeD study, had never been studied in this capacity and were absent from the literature.

This study examined parent perceptions at a point in time of the life course of their child with CHD that is less found in the literature, as most studies report parent outcomes during the pre-natal, peri-operative, or child's hospital discharge to home time periods (Bevilacqua et al., 2013; Brosig, Whitstone et al., 2007; Ezzat et al., 2016). This study also proved innovative by determining if mothers and fathers of a child with CHD from the same household were aligned in their perceptions of stress, QOL, and family functioning.

Study Purpose

The purpose of this descriptive, correlational, cross-sectional study was to determine how mothers and fathers of a child with CHD perceive their personal levels of stress, QOL, and family functioning, how these levels differ between parent pairs, how stress and QOL is related to family functioning, and how severity of infant CHD is related to parental stress, QOL, and family functioning. McCubbin and Patterson's (1983a, 1983b) double ABCX model of family adjustment and adaptation served as the theoretical framework for this study.

Research Questions

The research questions (RQ) and associated hypotheses in this study include:

RQ 1a: Among parents of a child with CHD, what is the difference in level of stress perceived by mothers and fathers?

Hypothesis 1a. Mothers of a child with CHD will report higher levels of stress compared to fathers.

RQ 1b: Among parents of a child with CHD, what is the difference in the perception of QOL between mothers and fathers?

Hypothesis 1b. Mothers of a child with CHD will report poorer QOL compared to fathers.

RQ 1c: Among parents of a child with CHD, what is the difference in the perception of family functioning between mothers and fathers?

Hypothesis 1c: Mothers of a child with CHD will report lower levels of family functioning compared to fathers.

RQ 2a: Among parents of a child with CHD, what associations exist between parental perceptions of personal stress and their family's functioning?

Hypothesis 2a: Parents who report high levels of stress will report low family functioning levels.

RQ 2b. Among parents of a child with CHD, what associations are present between parental perceptions of their QOL with their family's functioning?

Hypothesis 2b. Parents who report poor QOL will report low family functioning levels.

RQ 3a: What is the association between the severity of the child's CHD type (using STAT score as measurement) and their parents' stress level?

Hypothesis 3a: Parental stress levels will increase as the severity of CHD type increases.

RQ 3b: What is the association between the severity of CHD type (using STAT score as measurement) and parents' perceptions of their personal QOL?

Hypothesis 3b: Parents will report poorer QOL when their child has a more severe CHD type.

RQ 3c: What association exists between the severity of CHD type (using STAT score as measurement) and their parents' perceptions of family functioning?

Hypothesis 3c: Parents will report lower family functioning when their child has a more severe CHD type.

CHAPTER 2

SYSTEMATIC REVIEW OF LITERATURE[1]

Chapter 2 is a systematic review of the literature pertaining to quality of life for parents when having a child with a congenital heart defect (Gregory et al., 2018). The purpose of this systematic review was to identify how parental quality of life is affected when having a child with CHD. A systematic search of several databases yielded thirty-three quantitative cross-sectional or cohort studies that met inclusion criteria. This systematic review was published by *The Journal of Cardiovascular Nursing* on March 29, 2018; DOI:10.1097/JCN.0000000000000466. This review identified gaps in the literature that are addressed in the PinCHeD study.

Abstract

Background: As survival rates for infants born with severe forms of cardiac defects (congenital heart defect [CHD]) improve, attention is directed to evaluating factors that affect the child's short- and long-term outcomes including parental quality of life (QOL). Purpose: The purpose of this review was to identify how parental QOL is affected when having a child with a CHD. Factors that influence parental QOL when having a child with a CHD will also be described. Methods: A systematic search of CINAHL, EMBASE, PsycINFO, and PubMed databases was performed. Thirty-three quantitative cross-sectional or cohort studies were selected for inclusion and analyzed for quality reporting using

[1] This chapter is an article that was accepted for publication in the Journal of Cardiovascular Nursing. Accepted for publication November 2, 2017.

Strengthening the Reporting of Observational Studies in Epidemiology guidelines. Results: Heart defect severity, age of child, perceived support, and availability of economic resources were identified as factors affecting parental QOL. Parent gender was related to QOL and family functioning factors. Paternal outcomes were reported in 23 of the 33 studies (70%) with an average father participation rate of 40%. Conclusions: Having a child with CHD negatively affects parental QOL. Future research should include targeting fathers to improve understanding of their unique perceptions and needs. Longitudinal studies should also describe correlations of parental QOL with their child's developmental outcomes. Efficacy studies testing supportive interventions on outcomes such as improved adjustment and QOL are needed.

Introduction

Congenital heart disease (CHD) is one of the most common birth defects with approximately 40,000 infants born per year with CHD; 25% have a complex type with high mortality rates (Centers for Disease Control and Prevention, 2016; Oster et al., 2013). Survivors of CHD are at increased risk for neurodevelopmental differences caused by biological or environmental factors such as genetic differences, drug exposure, lengthy hospital stays, and psychological distress of parents (Brosig et al., 2014; Chock & Lee, 2014; Marino et al., 2012). Numerous studies focus on the major physical and psychosocial consequences of the CHD for the child with little attention to the relationships between the child's condition and their parent's quality of life (QOL) (Brown et al., 2005; Massaro et al., 2008). Relationships have been documented between a child's behavior, development, chronic condition, and vulnerability and a parent's feelings of stress (Bjarnason-Wehrens et al., 2007; De Ocampo, Macias, Saylor, & Katikaneni, 2003; Majnemer et al., 2006;

McCusker et al., 2007; Thomasgard & Metz, 1999). Parent and family factors may have greater effect on CHD child outcomes than the heart defect type or surgical palliation course (Rempel & Harrison, 2007).

The World Health Organization (1995) defines QOL as ‘‘the individual’s perception of his/her position in life in the context of culture and value systems in which he/she lives and in relation to his/her goals, expectations, standards, and concerns’’ (p. 1). Health-related QOL evolved from the QOL concept because of a growing consensus that QOL was too broad and did not capture the meaningful nuances on QOL that health influences (Haas, 1999). For concept identification in the scope of this systematic review, ‘‘QOL’’ and ‘‘health-related QOL’’ will be referred to as ‘‘QOL.’’

Systematic Review Methods

On September 1, 2016 PubMed (1946–September 2016), CINAHL (1981–September 2016), PsycINFO (1806–September 2016), and EMBASE (1947–2016) were searched using index terms from each database’s controlled vocabulary and truncated text words combined with Boolean connectors to form sets of citations on the search topic. Search terms used included *quality of life, QOL, health-related quality of life, HRQOL, psychological adaptation, coping, psychological stress, parents, parent, mother, father, congenital heart defects, abnormal heart, heart, family, and stress*. Full-length search strings used for each database are available from the first author upon request. This initial search generated 1306 articles that were analyzed further by title and/or abstract for relevance (N = 159). Remaining articles were uploaded into Endnote X7.7 and duplicates were removed resulting in 130 articles (see Figure 1) (Moher, Alessandro, Tetzlaff, & Altman, 2009). Inclusion criteria were: quantitative, cross-sectional or cohort design, sample of parents of a child with

CHD, self-reported parent QOL instruments used, full text in English, published in a peer-reviewed journal.

Data Extraction and Analysis

The Strengthening the Reporting of Observational Studies in Epidemiology (STROBE) (Vandenbroucke et al., 2007) statement is a guideline to assist researchers in addressing 22 key elements when writing cohort, case-control, and cross-sectional studies (von Elm et al., 2007). Separated into six sections: title and abstract, introduction, methods, results, discussion and other information, these elements, when reported, reveal "the strengths and weaknesses of a study and facilitates sound interpretation and application of study results" (Vandenbroucke et al., 2007). Items were scored using 0 (no information reported), 0.5 (partial information reported), or 1 (complete information reported). The STROBE total score range was 12-20 with a mean of 17.5. Two authors independently evaluated, extracted, and scored the included articles per the STROBE guidelines. Extraction accuracy and scoring was reviewed by a third author for agreement and accuracy against the original document if resolution in differing scores was needed. Individual author scores are available from the first author upon request (see Table 1).

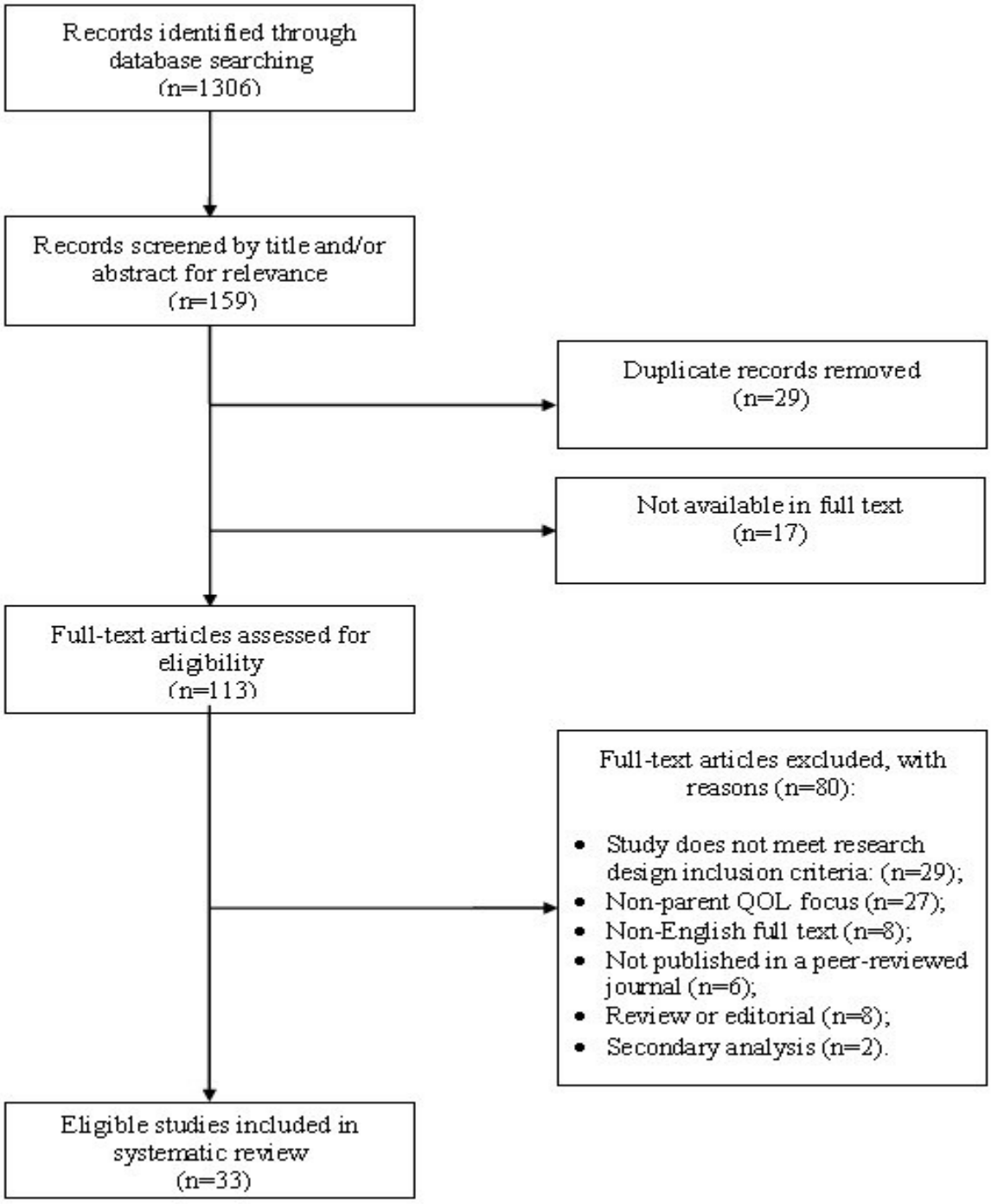

Figure 1. PRISMA Flow Chart for Study Selection (Moher et al., 2009).

Table 1

STROBE Quality Reporting Scores-By Section

	Title, abstract, and introduction section total	Methods section total	Results section total	Discussion section total	Other information section total	Total STROBE Score
Ahn, J., Lee, S., & Choi, J. Y. (2014)	3	7.5	4	2.5	1	18.0
Almesned, S. et al. (2013)	3	7	2.5	2	0	14.5
Arafa, M., Zaher, S., El-Dowaty, A., & Moneeb, D. (2008)	3	7.5	4	3.5	0	18.0
Bevilacqua, F. et al. (2013)	2.5	8	3.5	3	1	18.0
Brosig, C., Whitstone, B. et al. (2007)	2.5	6.5	3.5	4	1	17.5
Brosig, C., Mussatto, K. et al. (2007)	2.5	8	4.5	4	1	20.0
DeMaso, D. et al. (1991)	3	6	4.5	3	0	16.5
Diffin, J., Spence, Naranian, T., Badawi, N., & Johnston, L. (2016)	3	6.5	4	4	1	18.5
Doherty, N. et al. (2009)	2.5	7.5	3.5	3.5	1	18.0
Ezzat, S. et al. (2016)	3	8	3.5	3.5	1	19.0
Franck, L. et al. (2010)	2.5	7	3.5	4	1	18.0
Franich-Ray, C. et al. (2013)	2.5	5.5	4	3.5	1	16.5
Goldbeck, L., & Melches, J. (2005)	2.5	7	4	4	1	18.5
Grønning-Dale, M. et al. (2012)	2.5	8	4	3	1	18.5
Grønning-Dale, M. et al. (2013)	2.5	7.5	5	3	1	19.0
Hearps, S. et al. (2014)	2.5	7	4	3.5	1	18.0
Helfricht, S. (2008)	3	7	4	3	1	18.0
Jordan, B. et al. (2014)	2.5	6.5	3.5	4	1	17.5
Lawoko, S., & Soares, J. (2002)	3	7.5	3.5	4	0	18.0
Lawoko, S., & Soares, J. (2003b)	3	6.5	3.5	4	0	17.0
Lee, S., Yoo, H., & Yoo, J. (2007)	3	7	3.5	1.5	0	15.0

Table continues

	Title, abstract, and introduction section total	Methods section total	Results section total	Discussion section total	Other information section total	Total STROBE Score
Levert, E. M., Heilbing, W., Dulfer, K., van Domburg, R., & Utens, E. (2016)	2.5	6.5	4	3	1	17.0
Menahem, S., Poulakis, Z., & Prior, M. (2008).	1.5	4.5	4	3	1	14.0
Mörelius, E., Lundh, U., & Nelson, N. (2002).	3	6.5	5	2.5	0	17.0
Sarajuuri, A., Lonnqvist, T., Schmitt, F., Almqvist, F., & Jokinen, E. (2012).	2.5	6	2.5	4	1	16.0
Sira, N., Desai, P., Sullivan, K., & Hannon, D. (2014).	3	7.5	3.5	4	0	18.0
Spijkerboer, A. et al. (2007)	2.5	7.5	5	4	1	20.0
Svavarsdottir, E., & McCubbin, M. A. (1996)	3	7	3.5	4	1	18.5
Utens, E. et al. (2000)	2.5	6.5	5	3	1	18.0
Uzark, K., & Jones, K. (2003)	2	4.5	2.5	2.5	0	11.5
Visconti, K., Saudino, K., Rappaport, L., Newburger, J., & Bellinger, D. (2002)	3	7.5	4	4	1	19.5
Werner, H., Latal, B., Valsangiacomo Buechel, E., Beck, I., & Landolt, M. (2014)	3	7	5	3	1	19.0
Yildiz, A., Celebioglu, A., & Olgun, H. (2009).	3	7	4	2.5	0	16.5

Note. Vandenbroucke et al., 2007; von Elm et al., 2007

Two authors independently evaluated, extracted, and scored the included articles per the STROBE guidelines. Extraction accuracy and scoring were reviewed by a third author for agreement and accuracy against the original document if resolution in differing scores was needed. Individual author scores are available from the first author upon request (see Table 1). The following data were extracted: a) author/year, b) purpose, c) design, d) theory/model, e) setting, sample, country, parent characteristics, f) parental measures, and g) primary and secondary findings (see Table 2).

Table 2

Analysis of Included Articles

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Ahn et al. (2014) Cross-Sectional, Comparative STROBE total score: 18.0	To evaluate and compare adolescent patients' and their parents' coping strategies and knowledge of congenital heart disease.	Seoul, Korea-Outpatient pediatric cardiology clinic at a university-affiliated tertiary medical center *N* = 40 Parents of a child with congenital heart defect (PCCHD) 17.5% Fathers Sample age information: 50.0% age 39-45, 27.5% age 46-50, 22.5 % age 50-57. Mean age by gender not provided. Convenience sample 50 dyads eligible, 40 dyads participated PCCHD and their child with CHD	The Coping Inventory for Stressful Situations Leuven Knowledge Questionnaire for CHD	PCCHD reported use of task-oriented coping strategies most frequently (mean of 60.8, p<.0001). Mean scores for emotion-oriented and avoidance-oriented coping strategies were 46.93 and 48.48 (p<0.001) respectively. Gender-based scores are not provided in this study
Almesned et al. (2013)	To evaluate the financial,	Buriadah, Qassim, Saudi Arabia-Pediatric cardiology outpatient	Impact on family (IOF)	Families of children with complex CHD reported an overall mean of 61.3. Highest

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Cross-Sectional STROBE total score: 14.5	psychological, social, emotional impact associated with childhood CHD and the impact of the diagnosis on the children and their families in Saudi Arabia.	department at Prince Sultan Cardiac Center-Qassim *N* = 41 families, 21 families of child with complex CHD, 20 families with child with mild CHD % CHD Fathers: not provided Age information: not provided Convenience sample 41 families eligible, 41 families participated. Family groups divided by severity of their child's CHD (mild vs. complex)	scale-short version	impact was in the perceived Familial/Social Burden (mean = 23.29) followed by Mastery scale (mean = 18.00), Financial Burden (mean = 10.19), and Personal Strain (mean = 9.15). Scores for all domains were higher for families of child with complex CHD. Significant differences (P<0.05) were reported in the Family (p=0.000) and Mastery (p=0.000) domains. Gender-based scores are not provided in this study

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Arafa et al. (2008) Cross-sectional, Case-Control study STROBE total score: 18.0	To describe the health-related quality of life (HRQOL) of parents whose children are suffering from heart diseases and to identify the most important factors that could affect it.	Alexandria, Egypt-Two pediatric hospitals *N* = 400 PCCHD 10% Fathers Sample age information: Mean age and SD, 35.7, ±20.4. Mean age by gender not provided. Convenience sample PCCHD, Random selection of control group 400 eligible, 400 participated. Parents of children with minor illness	Structured questionnaire for SES, related heart disease, and family related risk data Health Survey-36 (SF-36)	In comparison to parents of children with minor illness, PCCHD had decreased vitality (39.66 vs 75.81), lower general health scores (46.25 vs 73.15), role limitations due to physical health (39.53 vs 61.81), decreases in physical functioning (75.76 vs 79.84), and decreases in social functioning (93.63 vs 98.88 (P<0.001). Financial situation affected physical and social functioning (F= 8.821, P<0.05 & F= 13.734, P<0.001) and was associated with lower vitality and emotional well-being scores. The mean score for parent overall QOL decreased as the child aged. Gender-based scores are not provided in this study

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Bevilacqua et al. (2013) Cross-sectional, Comparative STROBE total score: 18.0	To evaluate emotional distress, depression, and quality of life in parents of infants with severe CHD admitted for the first time to our children's hospital within first 3 months of life	Rome, Italy-Neonatal ward or cardiology unit at a Children's hospital *N* = 74 PCCHD 45% Fathers Mean age fathers: 36.4 Mean age mothers: 33.3 Convenience sample 76 eligible, 74 participated No control group	General Health Questionnaire (GHQ-30) (Italian version) Beck Depression Inventory, 2nd ed. (BDI-II)-Italian version Health Survey-36 (SF-36)	Mothers- 81.8% had significantly higher stress levels ($p<0.03$); 45.7% experienced depression; 13.8% experienced a difference in perceived mental health, 9.5% experienced a difference in perceived physical health. Fathers- 60.6% had significantly higher stress levels ($p<0.03$); 20% experienced depression, 12.2% experienced a difference in perceived mental health, .9% experienced a difference in perceived physical health.
Brosig, Whitstone et al. (2007) Mixed methods, Cohort, Comparative	To prospectively evaluate (at different time points) coping and psychological functioning of parents of children	Wisconsin, USA, Children's Hospital of Wisconsin *N* = 34 PCCHD 50% CHD Fathers Age information: not provided Convenience sample	Brief Symptom Inventory (BSI) Interview with semi-structured questions	BSI scores showed no significant difference between prenatal and postnatal parent groups at time of diagnosis (effect size= 0.26). At time of birth, 75% (*n* = 12) parents who received prenatal diagnoses had BSI scores in the clinically significant range. At six months after birth, prenatally diagnosed parent group had higher BSI

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
STROBE total score: 17.5	prenatally or postnatally diagnosed with CHD.	11 prenatally diagnosed families eligible, 10 (n = 20) participated; 16 postnatally diagnosed families eligible, 7 (n = 14) participated. Parent groups divided by timing of diagnosis of their child's CHD (prenatal vs postnatal) No control group		scores than postnatally diagnosed parent group (t=2.092, p=0.056, effect size= 0.42). Overall group means were not in the clinically significant range however, at time of diagnosis, 58% (n = 11) of the prenatally diagnosed parent group reported BSI scores in the clinically significant range compared to 71% (n = 10) of the postnatally diagnosed parent group. For the entire sample, there were no significance differences in the percentage of mothers versus fathers with clinically significant BSI scores at any time of data collection. χ^2-analyses demonstrated 81% of parents of a child with severe CHD had BSI scores in clinically significant ranges compared to 35% of parents of a child with less severe CHD.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
*Brosig, Mussatto et al. (2007b) * Companion article with Brosig, Whitstone et al. (2007) Cross-sectional, comparative STROBE total score: 20.0	To assess the psychosocial outcomes of preschool-aged survivors (ages 3–6 years) of hypoplastic left heart syndrome and transposition of the great arteries.	Wisconsin, USA, Children's Hospital of Wisconsin *N* = 26 PCCHD % CHD Fathers: not provided Age information: not provided Parents were divided by their child's CHD type only. Convenience sample 30 HLHS families eligible, parents of 13 HLHS families participated. 33 TGA families eligible, parents of 13 TGA families participated. Parents groups were divided by their child's CHD type (hypoplastic left heart syndrome (HLHS) vs transposition of the great arteries (TGA)). No control group	Impact on the family scale (IOF) Parenting stress index (PSI)	Entire CHD sample reported less negative impact on family functioning compared to families of children with other chronic illnesses ($p < 0.05$). HLHS parent group reported more negative impact on all subscales of family function compared to TGA parent group ($p < 0.05$ for all). Clinical meaningfulness is demonstrated by large effect sizes (range of 0.80-1.19) between parent groups for all subscales. Mean scores of parenting stress for PCCHD are significantly lower than normative values on total stress, the parent domain, and competence, attachment, role restriction, and depression subscales ($p<0.05$ for all). TGA parent group reported lower scores in all subscales when compared to test norms. HLHS parent group reported more negative impact on family functioning than TGA parent group in the subscales of child

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				demandingness, parental attachment, and depression ($p<0.05$ for all). Gender-based scores are not provided in this study.
DeMaso et al. (1991) Cross-sectional STROBE total score: 16.5	To determine whether severity of the heart disorder and maternal perceptions are related to the emotional adjustment of children with congenital heart disease.	USA-Outpatient cardiology clinic of a tertiary care pediatric hospital *N* = 99 PCCHD 0% Fathers Age information: not provided Convenience sample 104 eligible, 99 participated No control group	Parenting Stress Index (PSI) Parental Locus of Control Scale (PLOC) Perception of medical severity measure	Mean scores on PLOC, 90.78 (SD= 10.78) and PSI stress score, 8.48 (21.33), mean total score, 213.90 (SD= 36.53) were not significantly different from each scale's comparative, normed means. Gender-based scores are not provided in this study
Diffin et al. (2016) Cross-sectional, case-control,	To identify levels of NICU-related stress, and levels of psychological	Westmead, New South Wales, Australia-Children's Hospital *N* = 71 PCCHD 47.9% Fathers	Hospital Anxiety and Depression Scale; Coping Inventory for	Means scores for anxiety and depression were higher for PCCHD in comparison to the control group at all three time points becoming more closely matched over time.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
cohort The Double ABCX Model of Family Adjustment and Adaptation STROBE total score: 18.5	distress, reported by parents of infants admitted to the NICU for cardiac surgery	Mean age Fathers: 36 ±10 Mean age Mothers: 30 ±5 Convenience sample Cardiac parents eligible vs participated at Time point 1: 110, 71; Time point 2: 71, 51; Time point 3: 49, 49. Parents of heart healthy infants.	Stressful Situations; Family Support Scale (FSS); Parental Stressor Scale: NICU (PSS: NICU).	CHD fathers reported higher levels of social support than CHD mothers at time-point 1, 37.09 ± 11.66, 33.78 ± 9.40, respectively. CHD mothers had a higher mean score than CHD fathers on the 'Parental Role and Relationship', and 'Sights and Sounds' PSS: NICU subscales. Significant main effect of Parent Role (Mother/Father) on overall PSS: NICU scores, F (4, 63) = 4.24, p = 0.004; Pillai's Trace = 0.21. Significant effect of Parent Role (Mother/Father) on the 'Parental Role and Relationship' subscale, F (1, 69) = 9.98, p=0.002; mothers' scores were significantly higher than fathers' scores [t=−3.19, f=69, p=0.002] with a mean difference of−0.60, 95% CI (−0.97, −0.22). Fathers within each group reported higher levels of social support than mothers at time-point 1.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				PCCHD reported higher levels of social support than parents of heart-healthy children at the 12-month follow-up. Parents of heart-healthy children had higher levels of task-focused, and avoidance coping at each time-point in comparison to CHD parents. CHD fathers had the highest levels of emotion-focused coping activity at both time-point 1 and 12 months.
Doherty et al. (2009) Cross-sectional, comparative	To examine psychological functioning and coping styles in both mothers and fathers in the	Belfast, UK- Royal Belfast Hospital for Sick Children *N* = 140 PCCHD 50% CHD Fathers	Brief symptom index (BSI); Carver, Scheier, and Weintraub	33% CHD mothers and 18% of CHD fathers report global stress index (GSI) scores in the clinically significant ranges. CHD mothers showed scientifically significantly higher levels of psychopathology compared to CHD fathers (p= 0.011).

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
The Transactional Stress and Coping Model STROBE total score: 18.0	early months of life of their infant born with severe CHD	Mean age PCCHD: 32.65 Mean age CHD Fathers: 33.8 Mean age CHD Mothers: 31.5 Convenience sample, 73 families eligible, 70 families participated CHD mothers vs CHD fathers	multi-dimensional coping inventory (COPE)-Situational version Townsend score Maternal worry Scale Significant others scale Family environment scale	CHD mothers reported statistically significant ($p<0.05$) higher mean scores compared to CHD fathers in the following COPE subscales: Instrumental ($p=0.02$) and emotional ($p=0.001$) social support, and religion ($p=0.03$), and venting ($p=<0.001$). CHD fathers reported alcohol use significantly more than CHD mothers ($p=0.01$). All PCCHD had significant associations between their mental health and family personal factors of knowledge and appraisal (CHD mothers $p=.003$, CHD fathers $p=0.006$), coping (CHD mothers-denial behavioral disengagement and disposition maladaptive, $p<0.001$-0.01; CHD fathers-mental disengagement, alcohol use, and humour, $p<0.001$-0.04) and, conflict (CHD mothers $p<0.001$, CHD fathers $p=0.018$).

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Ezzat et al. (2016) Mixed Methods: Cross-sectional, Case-Control study Grounded theory approach was used to ensure rigorous analysis STROBE total score: 19.0	To determine the perceived causal attributions of disease among parents of children with congenital cardiovascular malformations; to determine the relationship of these attitudes and perceptions to time to diagnosis; to assess stress among mothers and its relationship to their knowledge of and health beliefs about	Cairo, Egypt-Hospital setting *N* = 99 CHD mothers 0 % Fathers Mean age Fathers: n/a Mean age Mothers: 28.6 years (SD=6.0, range of 18-55). Purposefully selected sample for cases, recruited sample for controls, Unknown # eligible, 99 CHD mothers participated. Mothers of heart healthy children	Semi-structured questionnaire Parent health locus of control scale Knowledge of heart disease and its treatment Parenting stress index - short form (PSI-SF) Religiosity questions	Non-CHD mothers had statistically significant higher scores on total stress and all subscales in comparison to CHD mothers: Total stress 92.8, 86.2 ($p<0.001$), Parental distress: 78.9, 72.3 ($p<0.05$); Parent-child dysfunctional interaction: 97.5, 93.0 ($p<0.01$), Difficult child: 73.0, 58.9 ($p<0.001$). Overall, all mothers had extremely high levels of stress (88.8 %, SD=15.1), $p<0.001$). CHD mothers felt more empowered to control their child's health than did non-CHD mothers ($t(162) = 2.24$, $p<0.05$). CHD and non-CHD mothers reported different significant relationships between difficult child and locus of control. Maternal stress was not related to religiosity (data not provided in study).

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
	congenital cardiovascular malformation in their children; to assess knowledge of congenital cardiovascular malformations and its relationship to parental health beliefs; and to assess religiosity among mothers and its relationship to stress and health beliefs			Gender-based scores are not provided in this study.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Franck et al. (2010) Prospective Case-cohort, comparative STROBE total score: 18.0	To investigate pre- and post-operative parental stress and to examine some of the influencing factors during the postoperative period for children undergoing elective cardiac surgery.	England- a large London children's hospital *N* = 211 PCCHD % CHD Fathers: not provided Age information: not provided Convenience sample 274 PCCHD eligible, 231 PCCHD participated Comparing PCCHD stress levels at pre-operative and post-operative days 3, 5, 8, and 15. No control group	Parent stressor scale: Infant hospitalization (PSS–IH) 4-additional items assessing parent's perspective on their child's overall health, their expectations of their child's recovery, and quality of pre-operative information received.	Stress scores for PCCHD (both genders) were correlated at all time points (r = 0.52–0.95). CHD fathers had lower stress scores than CHD mothers for three time points (preoperatively, day 3, and day 5; $p<0.05$). At all time points, PCCHD had highest stress scores for these subscales: child behavior & appearance, parental role, and sights & sounds. Mothers' ratings of child health were correlated with their PSS-IC score for postoperative day 5 (rho = –.27; $p<0.05$). There was no correlation between fathers' ratings of child health and their PSS-IC scores.
Franich-Ray et al. (2013)	To investigate the prevalence	Melbourne, Australia-Royal Children's Hospital	Acute stress disorder scale	27% of all parents met criteria for acute stress disorder.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Cross-sectional STROBE total score: 16.5	and nature of trauma symptoms in mothers and fathers of infants who had cardiac surgery before 3 months of age.	*N* = 132 PCCHD 41.6 % Fathers Mean age Fathers: 35.5 (SD=5.5), range 23.2-48.9 Mean age Mothers: 32.9 (SD=4.9), range= 19.9-42.0 Convenience sample 176 parents eligible, 132 participated No control group		CHD Mothers- 33.8% met criteria for acute stress disorder. Symptom cluster mean scores: Dissociative: 12.84; Re-experiencing: 7.91, Avoidance: 7.69, Arousal: 10.95, Overall: 39.42. CHD Fathers- 18.2% met criteria for acute stress disorder. Symptom cluster mean scores: Dissociative: 11.36, Re-experiencing: 6.60, Avoidance: 6.44, Arousal: 9.20, Overall: 33.60. Timing of CHD diagnosis (pre- vs. post-natal) significantly impacted likelihood of experiencing at least one arousal symptom in fathers, of at least one avoidance symptom at a clinical level in parents.

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Goldbeck and Melches (2005) Cross-sectional, comparative STROBE total score: 18.5	To explore the extent of agreement on the patient's QOL between children and adolescents with CHD and their caregivers, to explore the association of the caregivers' own QOL with their children's QOL, to explore the degree of moderation parental QOL has on parent-child agreement on	Germany-Outpatient pediatric university CHD clinic *N* = 69 PCCHD 16% CHD Fathers (*n* = 11) Age information: not provided Convenience sample. 180 CHD families eligible, 143 CHD families participated. PCCHD and their child with CHD	Ulm quality of life inventory for parents (ULQIE)	The majority of all participants reported positive perceptions of QOL with median scores ranging from 75-92 and means ranging from 78.6-88.6. Statistically significant correlations between parent proxy reports and child self-report for the following items: Psychological well-being/ function ($r=0.61$, $p<0.001$), disease and therapy-related distress ($r= 0.56$, $p<0.001$), physical well-being/ function ($r=0.51$, $p<0.001$). Significant general interaction effect of parental QOL and children's self-rated QOL ($t = 3.61$; $p < 0.001$). Conditional regression indicates PCCHD with low QOL scores agree better with their CHD child's self-perception than parents with high QOL.

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
	the patients' QOL.			
*Grønning-Dale et al. (2012) *Companion article with Grønning-Dale et al. (2013) Prospective, case-cohort, comparative STROBE total score: 18.5	To compare the well-being among mothers of children with congenital heart defects (CHD) with mothers of children without CHD (controls), at pregnancy and at 6 months postpartum	Oslo, Norway-Department of Pediatrics, Pediatric Cardiology Unit, at Rikshospitalet University Hospital *N* = 212 CHD mothers 0% CHD Fathers Mean age of mothers at time of child's birth: Control= 30.1 ± 4.52, mild CHD 30.7 ± 4.31, moderate CHD 30.4 ± 4.30, severe CHD 29.8 ± 4.29 Convenience sample 252 CHD mothers eligible, 212 CHD mothers participated CHD mothers grouped by CHD severity: Mild CHD, *n* = 92,	Satisfaction with life scale (SWLS) Differential emotions scale (DES), joy and anger subscales Social support questionnaire	CHD severity did not have statistically significant main effect on maternal life satisfaction [F (3, 57,442) = 1.945. p= 0.120] CHD mothers' feelings of joy were generally similar to the findings of their life satisfaction. The overall effects of CHD severity on feelings of joy were not significant. The effect of time on mothers' feelings of anger was dependent on the severity of CHD at 6 months postpartum. At 6 months post-partum, severe CHD mothers had significantly higher anger scores than controls (standard deviation = 0.34)

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
		Moderate CHD, *n* = 50, Severe CHD, *n* = 70 Mothers of children without CHD		
Grønning-Dale et al. (2013) Prospective case-cohort, comparative STROBE total score: 19.0	To explore the effects of and relationships with congenital heart defects (CHD) on mothers' well-being over time.	Oslo, Norway-Department of Pediatrics, Pediatric Cardiology Unit, at Rikshospitalet University Hospital *N* = 175 CHD mothers 0% CHD Fathers Mean age of mothers at time of child's birth: Control= 30.3 ± 4.45, mild CHD 30.6 ± 4.93, moderate CHD 30.6 ± 4.56, severe CHD 30.1 ± 4.22 Convenience sample 252 CHD mothers eligible, 175 CHD mothers eligible CHD mothers grouped by CHD severity: Mild CHD, *n* = 79,	Satisfaction with life scale (SWLS) Differential emotions scale (DES), joy and anger subscales	Overall effects of CHD severity on maternal SWB were significant [F(3, 41649)=3.649, p =0.012] with severe CHD mothers having significantly lower overall scores on social well-being (SWB) than controls (p=0.012). Overall effects of time on maternal life satisfaction were significant [F(2, 83298)=24.064, p<0.0001] and all CHD groups had significantly lower overall SWB scores at 36 months postpartum compared to SWB scores prenatally (p<0.0001) and at 6 months post-partum (p<0.0001) indicating time is an important factor in explaining changes in mothers' well-being. Significant interaction effect between time period and CHD group [F(6, 83298)=2.406, p=0.025]. Severe CHD mothers had the lower SWB scores compared to controls at

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
		Moderate CHD, *n* = 36, Severe CHD, *n* = 60) Mothers of children without CHD		all time points with a further decrease in SWB at 36 months post-partum. Gender-based scores are not provided in this study.
Hearps et al. (2014) Cross-sectional, comparative STROBE total score: 18.0	To investigate the frequency and nature of parent psychosocial risk occurring after surgery for congenital heart disease and the impact of the time of diagnosis, antenatal or postnatal.	Melbourne, Australia-Royal Children's Hospital *N* = 39 PCCHD 28.2% Fathers Mean age Fathers: not provided Mean age Mothers: 20–29 years range, *n* = 13, 33.3% 30–39 years range, *n* = 23, 59.0% 40+ years range, *n* = 2, 5.1% Convenience sample 68 eligible, 39 parents participated No Control Group	Psychosocial assessment tool (PAT)	38% PCCHD self-rated as experiencing psychosocial risk in the clinical (high) or targeted (medium) ranges. Scaled scores for PAT subscales (Scaled range= 0-1): Structure/resources: 0.12 Social support: 0.06 Child problems: 0.19 Sibling problems: 0.07 Family problems: 0.13 Stress reaction: 0.14 Family beliefs: 0.10 Parental education level was the sole significant predictor of the total PAT score, with those having high school education less recording a significantly higher PAT than those with at least some tertiary education (β= -0.85, p=0.005).

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				No significant correlation was found between mothers and fathers of same families on the PAT total score (dyad *n* = 10, r=0.59, p>0.05).
Helfricht et al. (2008) Prospective cohort STROBE total score: 18.0	To contrast paternal levels of surgery-related PTSD symptoms with PTSD symptoms in mothers. To explore risk factors relating to pre-, peri-, and postoperative data of the child for surgery-related PTSD in parents.	Zurich, Switzerland- University Children's Hospital *N* = 233 PCCHD 42% Fathers Mean age Fathers: 37.5 (SD= 6.7) Mean age Mothers: 34.6 (SD=5.5) Convenience sample 228 families eligible, 139 families participated No Control Group	The posttraumatic diagnostic scale (PDS)	No statistically significant gender difference for rates of PTSD. Severity of PTSD symptoms declined significantly in mothers and fathers between assessments (t= 7.11, p<0.01) Fathers, t= 6.49, p< 0.01). PTSD severity post-discharge correlated with PTSD severity at 6 months in all parents.
Jordan et al. (2014)	To explore mothers'	Australia	Maternal postnatal	No difference in attachment feelings between CHD mother's and community

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Mixed Methods, Cross-sectional STROBE total score: 17.5	subjective experiences of the relationship with their infant soon after discharge from hospital post-cardiac surgery and describe the impact of medical variables or maternal depression on the mother-baby relationship.	*N* = 91 CHD mothers 0 % Fathers Mean age Fathers: n/a Mean age Mothers: 32.9 (SD=4.9) Convenience sample 115 eligible, 97 participated Australian community norms	attachment scale (MPAS) Edinburgh postnatal depression scale (EPDS) Questionnaires and interviews	norms per MPAS. However, qualitative responses of CHD mothers demonstrated increased maternal protectiveness and care. Almost 25% CHD mothers reported difficulty bonding with their infant. 20% CHD mothers reported anxiety, stress, and fear dominated the maternal-infant relationship which was associated with prenatal diagnosis. Gender-based scores are not provided in this study
*Lawoko and Soares (2002) *Companion article with Lawoko and	To examine differences in symptoms of depression, anxiety, and somatization,	Sweden *N* = 1497 (1092=PCCHD, 112=PCOD (Parent of child with other disease), 293=PHC (Parent of healthy child))	The symptom checklist-revised (SCL-90-R) Hopelessness Scale	CHD mothers devoted about 2 hours extra time to caring for their sick children in contrast to 1/2 hour for CHD fathers [t(1079) = 3.21,P < .01].

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Soares (2003b) Cross-sectional, comparative STROBE total score: 18.0	and hopelessness between parents of a child with CHD (PCCHD), parents of a child with other diseases (PCOD), parents of healthy children PHC).	40% all fathers, 39% CHD fathers Mean age CHD Parents: 39 ±7 Convenience sample of PCCHD, random selection of comparative sample, 1500 PCCHD eligible, 1092 PCCHD participated. PCOD, PHC, and psychiatric outpatient norms (POPN).		CHD mothers had higher global stress index scores than COD mothers ($p<0.05$) and HC mothers ($p<0.005$). CHD fathers had higher global stress index scores than HC fathers ($p<0.05$). PCCHD had higher depression and anxiety scores than PCOD ($p<0.005$) and PHC ($p<0.001$). PCCHD and PCOD were more worried about their financial situation than PHC [$x^2(2) = 10$, $p<0.01$]. In all groups, mothers had higher GSI scores than fathers [PCCHD, $t(1086) = 8.2$, $p<0.001$; PCOD, $t(108) = 2.6$, $p<0.05$; PHC, $t(286) = 3.3$, $p<0.005$]. In all groups, mothers had higher depression scores than fathers [PCCHD, $t(1086) = 9.1$, $p<0.001$; PCOD, $t(108) = 2.3$, $p<0.05$; PHC, $t(286) = 3.7$, $p<0.001$].

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				Both CHD mothers and CHD fathers have higher hopelessness scores than mothers and fathers of healthy children ($p<0.005$ and $p<0.05$). In all groups, mothers had higher anxiety-scores than fathers [PCCHD, $t(1086) = 6.9$, $p<0.001$; PCOD, $t(108) =2.4$, $p<0.05$; PHC, $t(286) = 2.8$, $p<0.01$]. In all groups, mothers had higher somatization scores than fathers [PCCHD, $t(1086) = 5.8$, $p<0.001$; PCOD, $t(108) = 2.3$, $p< 0.05$; PHC, $t(286) = 2.6$, $p<0.05$].
Lawoko and Soares (2003b) Cross-sectional; comparative	To compare social support experiences of parents with children who have CHD to parents with children who have other diseases and	Sweden *N* = 1497 (1092=PCCHD, 112=PCOD, 293=PHC) 39% CHD Fathers Mean age CHD Parents: 39 ±7	The schedule for social interaction The symptom checklist-revised (SCL-90-R) The	Child's health did not independently explain social support availability to CHD parents. Financial instability (9%) explained the most variability among social support in CHD parents, children's variables explained 2%, parent demographics, 3%, and elevated psychological distress & hopelessness, combined, accounted for 7% of the

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
STROBE total score: 17.0	parents who have healthy children.	Convenience sample PCCHD, Random selection of comparative sample, 1500 PCCHD eligible, 1092 PCCHD participated Parents of child with other diseases (PCOD) and Parents of healthy children (PHC)	Hopelessness scale Three structured, finances-based questions	variation of available social support for CHD parents. Statistically significant correlations were found between social interaction and social integration and anxiety, depression, somatization, global severity index and hopelessness for CHD parents (ranges of +0.2 to +0.5 and -0.3 to -0.4, $p < 0.01$). CHD mothers had lowest availability of social support of all parent groups. CHD mothers spend more time caring for their ill child than CHD fathers. Fathers- All fathers reported greater availability of social interactions.
Lee et al. (2007) Cross-sectional	To examine the relationships among uncertainty, social support, and parenting stress in CHD mothers and to	Seoul, Korea-Pediatric cardiac outpatient clinic *N* = 51 PCCHD 0% Fathers Mean age Fathers: n/a	Personal resource questionnaire (PRQ) Parenting stress index-	Social support and information obtained from the internet were significant determinants and together accounted for 39.4% of parenting stress (p=0.032). Parenting stress was significantly related to social support, ambiguity, lack of clarity and

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
STROBE total score: 15.0	identify factors related to parenting stress.	Mean age Mothers: 20–30 range, *n* = 8, 15.7% 31–40 range, *n* = 37, 72.5% > 40 range, *n* = 6, 11.8% Convenience sample Unknown # eligible, 51 participated No control group	short form (PSI/SF) Parent's perception uncertainty in illness scale (PPUS)	lack of information but NOT unpredictability (p<0.01). CHD mothers' parenting stress was significantly related to their child's age (p<0.01), and mother's education level (p=0.03) but NOT the child's cardiac defect type. Gender-based scores are not provided in this study.
Levert et al. (2016) Cross-sectional STROBE total score: 17.0	To investigate the psychosocial needs of both parents of children with CHD (aged 0–18 years) and patients themselves (aged 8–18 years) in the week before cardiac surgery	Netherlands *N* = 161 PCCHD 52.7% Fathers Age information: not provided Convenience sample 282 eligible, 161 participated	Online disease-specific questionnaire designed for this study Linear analogue scale	In general, majority of PCCHD reported an increased need for psychosocial care for themselves when their children were aged 0–12 years in the domains of Physical/Medical, Emotional well-being, social, educational/occupational, social support, and Health behavior (p=0.10). Parents of children with complex CHD demonstrated greater need for care in all domains (p≤0.20) Gender-based scores are not provided in this study.

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
	or a catheter intervention.			
Menahem et al. (2008b) Prospective Cohort STROBE total score: 14.0	To investigate the psychological and emotional experiences of parents when their children are subjected to cardiac surgery.	Australia *N* = 57 PCCHD 49 % Fathers Age information: not provided Convenience sample 206 eligible, 57 participated No control group	State-trait anxiety inventory; General health questionnaire (GHQ); Levenson's locus of control questionnaire Family assessment device Index of social support	Substantial increase in CHD mothers' emotional distress at the time of their child's cardiac surgery resolved by 12 months or later (Reduction of means= 16.13, p=0.0001). The 'lack of control in their life' feeling reported by PCCHD persisted beyond 12 months for unclear reasons. Gender-based scores are not provided in this study. NOTE: Authors report that most data was obtained from mothers; only 10 fathers completed the questionnaires.

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Mörelius et al. (2002) Retrospective, cross-sectional, comparative STROBE total score: 17.0	To clarify whether differences exist in parental stress when the child has a complex CHD compared to a minor CHD.	Sweden *N* = 101 parents 44.5 % Fathers Age information: not provided Convenience sample 144 eligible, 101 participated. Parent groups divided by severity of their child's CHD (minor vs. complex)	Parenting stress index (SPSQ), Swedish version	CHD severity has no statistical difference between complex CHD total SPSQ (mean =85.2, SD= 16.7, p=ns) and minor CHD total SPSQ (mean = 82.5, SD=15.6, p=ns). There were no significant differences between CHD severity groups in any subscale for mothers or for fathers.
Sarajuuri et al. (2012) Cross-sectional, comparative STROBE total score: 16.0	To assess perceptions of child behaviour and parenting stress among the parents of young children with hypoplastic left heart syndrome	Finland, Children's Hospital of Helsinki University Central Hospital *N* = 83; parents of 23 children with HLHS, parents of 14 UVH children, parents of 46 healthy children 73 mothers and 2 (2%) fathers completed the CBCL.	Parenting stress index (PSI) Child behavior checklist (CBCL)	HLHS mothers (mean score 241 vs 205, p<0.001) and HLHS fathers (mean score 235 vs 202, p=0.003) reported significantly higher total parenting stress scores than same-gender controls. HLHS parents reported significantly more total (mean T score 52 vs 45, p=0.005) and internalizing behavior problems (51 vs 41, p<0.001) than controls.

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
	(HLHS) and other forms of functionally univentricular heart defects (UVH).	81 mothers and 54 (65%) fathers completed the PSI. Median (range) age of mothers at birth: HLHS 29 (18-40), UVH 30 (17-40), Control 31 (19-42) Convenience sample Parents of 29 children with HLHS and 23 children with UVH were eligible; parents of 23 children with HLHS and 14 children with UVH participated. HLHS parents, UVH parents of child, and PHC.		There was a statistically significant ($p=0.007$) difference in somatic complaints between HLHS parents and controls. On the PSI: Isolation subscale- Significant difference between CHD mothers and mothers of healthy children (13 vs 14, $p=0.037$). Competence subscale- Significant differences were reported between HLHS mothers and mothers of healthy children (31 vs 25.5, $p=0.0001$) and HLHS fathers and fathers of healthy children (29 vs 25, $p<0.05$). Role restriction subscale- Significant differences were reported between HLHS mothers and mothers of healthy children (22 vs 18.5, $p<0.05$); Depression subscale- Significant differences were reported between HLHS mothers and mothers of healthy children (24 vs 18.5, $p<0.01$). Health subscale-There was a significant difference between CHD mothers and mothers of healthy children (14 vs 11.5,

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				p=0.027) and CHD fathers compared to fathers of healthy children (12 vs 11.5, p=0.034).
Sira et al. (2014) Cross-sectional The double ABCX model of adjustment and adaptation STROBE total score: 18.0	To explore internet use as a coping resource for parents of children with CHD.	USA *N* = 175 (178 mothers, 3 fathers) 0% Fathers (these 3 responses were omitted from this study) Sample age information: 15.3% age 21-30; 65% age 31-45; 19.3 % age 46-60. Mean age by gender not provided. Convenience sample Unknown # eligible, 178 participated. No control group	Coping health inventory for parents (CHIP) Spiritual health and behavioral scale (SIBS) Internet use survey 3, open-ended questions	41.1% scored high in coping pattern I, which measures family integration maintenance, cooperation, optimism behaviors. 23.4% scores high on Coping Pattern II which involved a mother's effort to maintain individual self-esteem and psychological stability. 83.4% had high scores for Coping Pattern III which includes effect in medical communication and consultation. 38.9% ranked spirituality as a high importance for coping. 69.7% mothers reported seeking medical information via the internet very often. CHD Fathers- due to low response rate (*n* = 3), this data was omitted from the study.

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Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Spijkerboer et al. (2007) Cross-sectional STROBE total score: 20.0	To assess the level of psychological distress and styles of coping in both mothers and fathers of children who underwent invasive treatment for congenital cardiac disease at least 7 years and 6 months ago.	The Netherlands-Erasmus University Medical Centre Rotterdam *N* = 161 PCCHD 38% CHD Fathers Mean age CHD mothers that completed the Utrecht coping list (UCL) 40.5±4.8; the GHQ, 40.5±4.7 Mean age CHD fathers that completed the UCL 44.6±6.0; the GHQ, 44.5±5.7 Convenience sample 159 patients eligible, parents of 109 patients participated No control group	The General health questionnaire (GHQ) Utrecht coping list (UCL)	PCCHD reported less complaints than normative sample on the GHQ's total score, somatic symptoms, anxiety & sleeplessness, and serious depression subscales ($p<0.05$). PCCHD of girls reported significantly more complaints on the serious depression scale than CHD parents of boys ($p<0.05$). CHD mothers reported significantly more somatic symptoms and higher when asked about the search for social support than CHD fathers ($p<0.05$). PCCHD had coping style tendencies that differed statistically compared to their same-gendered norms ($p<0.05$). Socioecomonic status showed a significant main effect on the variance of all GHQ scales. CHD mothers showed significantly higher tendency to seek social support as a coping means than CHD fathers ($p<0.05$).

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Svavarsdottir and McCubbin (1996) Cross-sectional, correlational The Resiliency Model of Family Stress, Adjustment, and Adaptation. STROBE total score: 18.5	To examine relationships between care giving demands, family system demands, and parental coping behavior in families with an infant (0-12 months of age) diagnosed with CHD	Midwest, USA-Participant homes *N* = 142 PCCHD 50% Fathers Mean age Fathers: 30.6 Mean age Mothers: 28.7 Convenience sample Unknown # eligible, 142 participated. No control group	The family profile inventory Family inventory of life events (FILE) Child illness factors scale (10-items) The coping health inventory for parents (CHIP) The care of	CHD mothers-infant feeding was reported as the most time-consuming task (mean = 3.47); providing emotional support for spouse or partner as the most difficult care-giving task (mean = 2.44). Younger mothers reported more helpful coping related to strengthening family life and maintaining optimism (r=-0.25, p<.05). Infant's illness severity positively correlated with understanding the health care situation (r=.28, p<.05). CHD fathers- designated providing emotional support for the spouse or partner as the most time-consuming task (mean = 3.20) and the most difficult (mean = 2.49). CHD fathers who had been married or in the relationship longer (r=-0.24, p<.05), or had

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
			my child measure (a modification of the Caregiving burden scale)	more children (r=-0.27, p<.05) reported less helpful coping behaviors related to social support, self-esteem, and psychological stability.
Utens et al. (2000) Cross-sectional, comparative STROBE total score: 18.0	To assess the level of psychological distress and the styles of coping of parents of children with CHD. To compare the same parameters in mothers and fathers of children awaiting surgery to those of	Netherlands-University Hospital of Rotterdam *N* = 206 PCCHD 49.5% Fathers Mean age Fathers of children awaiting cardiac surgery: 34.3, SD=4.9 Mean age Fathers of children awaiting cardiac catheterization: 34.7, SD= 4.4 Mean age Mothers of children awaiting cardiac surgery: 31.5, SD= 5.5 Mean age Mothers of children awaiting cardiac catheterization: 32.2, SD= 4.1	General health questionnaire (GHQ) The Utrecht coping list	All parents (regardless of gender) of children awaiting surgery reported to express their anger or annoyance to a lesser extent in comparison to reference sample. Mothers scored significantly higher than fathers in terms of somatic symptoms and anxiety or sleeplessness. For the most part, there were no significant differences between parents of children scheduled for surgical correction versus interventional catheterization. CHD mothers- reported significantly more complaints on the total score of the GHQ than reference sample (7.57, p<0.05).

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
	mothers and fathers of children awaiting interventional catheterizations for CHD.	Convenience sample 256 eligible, 206 participated. Parents of children having cardiac surgery/ parents of children having cardiac catheterization procedure		CHD fathers- reported significantly more complaints regarding social dysfunctioning than reference sample (7.97, $p<0.05$).
Uzark and Jones (2003) Cross-sectional Abidin's Parenting Stress Index Model STROBE total score: 11.5	To examine parenting stress reported by parents of children older than 2 with CHD.	Cincinnati, Ohio, USA-Pediatric outpatient cardiology clinic *N* = 80 PCCHD 12.5% Fathers Age information: not provided Convenience sample Unknown # eligible, 80 participated. No control group	Hollingshead (1975) four-factor index measured SES Parenting stress index-short form (PSI-SF)	Parent stress is unrelated to severity of CHD, or time since most recent surgery (r-0.190, p= not significant). PCCHD reported parent-related stress in excess of that expected on the basis of normative data. Gender-based scores are not provided in this study.
Visconti et al. (2002)	To examine the role of parent stress and social support	Boston, Massachusetts, USA-Boston Children's Hospital *N* = 277 PCCHD	Parent stress index-PSI Child	PCCHD reported significantly less parent-related stress at 1 and 4 years of child's age (mean = 115.5 and 153. $p<0.001$) than normative sample (mean = 122.7).

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
Prospective, cross-sectional, comparative STROBE total score: 19.5	in the emotional adjustment of children with d-transposition of the great arteries (d-TGA).	48% Fathers Age information: not provided Convenience sample 163 eligible families, 143 and 145 participated at 1 and 4 years of child's age respectfully. No control group	behavior checklist (CBCL) Hollingshead four factor index of social status Social support network inventory (SSNI)	PCCHD perceived significantly fewer internalizing behavior problems in their child than normative sample (mean = 46.5, p=0.0001). PCCHD that reported less perceived social support experience more stress at 1 & 4 years of child's age. (-0.43, p≤0.001, -0.41, p≤0.001). Parent stress and child's problem behavior were positively correlated on multiple subscales. Parents who reported more social support tended to have children with lower total problem behavior and externalizing scores on the CBCL. At 1 year of age, families with high levels of social support reported more behavior problems than families with low levels of social support. Social support did not appear to buffer effects of stress on child

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				adjustment. Gender-based scores are not provided in this study.
Werner et al. (2014) Prospective, cohort, comparative STROBE total score: 19.0	To investigate the impact of a child's severe CHD on the family and to prospectively examining the influence of disease specific and psychosocial factors on the family.	Zurich, Switzerland-University Children's Hospital *N* = 147 PCCHD 45% Fathers Mean age Fathers: 36.2 Mean age Mothers: 34.1 Recruited convenience sample, 192 eligible, 147 participated. No control group	Impact on family scale-Generic (IOF-G), German version Social support questionnaire - Short form (F-SozU-K-14) Demographic questionnaire	Families with poor social support network may have greatest need for professional interventions. No significant difference was identified for the IOF between mothers and fathers (z= -0.69, p= 0.49). Underlying genetic defect, long hospital stays, and lower levels of social support were attributed as having greatest impact on family. CHD mothers- Higher reports of having to live with more ups and downs than fathers (Cohen's d = 0.30) CHD fathers- Higher reports of having to give up things and seeing family members

Table continues

Author & Year; Study Design Theory/Model used (if any); Total STROBE quality score	Purpose	Setting and Sample Characteristics (N, % male, CHD parent age data, Eligible/participated rate); Comparative/Control group(s)	Parental QOL / family functioning measures used	Primary Findings: QOL / family functioning factors; Secondary Findings: Significant gender differences
				and friends less frequently than mothers (Cohen's d= 0.27 & 0.30, respectively).
Yildiz et al. (2009) Cross-sectional, comparative STROBE total score: 16.5	To determine the distress levels of parents of children with CHD and identify factors that influences the levels of stress.	Erzurum, Turkey-Pediatric cardiology outpatient clinic *N* = 262 PCCHD 49.6% Fathers Mean age Fathers: 20–29 age range: *n* = 17. 13.1% 30–39 age range: *n* = 77, 59.2% 40+ age range: *n* = 36, 27.7% Mean age Mothers: 20–29 age range: *n* = 59, 44.7% 30–39 age range: *n* = 56, 42.4% 40+ age range: *n* = 17, 12.9% Convenience sample Unknown # eligible, 262 participated. No control group	Researcher composed, closed-ended questionnaire Symptom checklist-90-Revised (SCL-90-R) Brief symptom index (BSI) Global severity index (GSI)	CHD mothers scored statistically significantly higher ($p<0.001$) than CHD fathers on all distress dimensions (somatization (1.17±0.43), anxiety (1.78±0.52), depression (1.54±0.50), and GSI (1.48±0.43). CHD mothers were more affected by their children's disease than CHD fathers, possibly because mothers are more actively engaged in their children's care than fathers, more often in communication with their children and spend much more time with their children. Although not always statistically significant, parent SCL scores were higher with CHD severity.

Table continues

Results

Research Design

The 33 included studies represent a total of 5,794 parents of a child with CHD. In the situation of companion publications, only the publication with the largest sample size was counted in the parents of a child with CHD sample total. Designs were cross-sectional (N = 25) or cohort (N = 8), and all contained quantitative measures of parental QOL. Studies used varied categories to group participants for comparisons including: timing of CHD diagnosis or surgical intervention (N = 6) (Brosig, Whitstone et al., 2007; Franck et al., 2010; Grønning-Dale et al., 2012; Hearps et al., 2014; Helfricht et al., 2008), CHD type or severity (N = 5) (Almesned et al., 2013; Brosig, Mussatto et al., 2007; Mörelius et al., 2002; Sarajuuri et al., 2012; Utens et al., 2000), parent and their adolescent child with CHD (N = 3) (Ahn et al., 2014; Goldbeck & Melches, 2005; Levert, Heilbing, Dulfer, van Domburg, & Utens, 2016), CHD mothers and CHD fathers (N = 10) (Bevilacqua et al., 2013; Diffin et al., 2016; Doherty et al., 2009; Franich-Ray et al., 2013; Helfricht et al., 2008; Spijkerboer et al., 2007; Svavarsdottir & McCubbin, 1996; Utens et al., 2000; Werner et al., 2014; Yildiz et al., 2009), parents of healthy children (N = 7) (Diffin et al., 2016; Ezzat et al., 2016; Grønning-Dale et al., 2012; Grønning-Dale et al., 2013; Lawoko & Soares, 2002, 2003b; Sarajuuri et al., 2012), parents of children with other diseases (N = 3) (Arafa et al., 2008; Lawoko & Soares, 2002, 2003b), or psychiatric outpatients (N = 1) (Lawoko & Soares, 2002). Parent ages ranged from 18–57 years. Studies were published between 1991 (DeMaso et al., 1991) and 2016 (Diffin et al., 2016; Ezzat et al., 2016; Levert et al., 2016) (see Table 2).

Setting and Sample

The 33 articles included studies occurring in 15 countries: United States ($N = 7$) (Brosig, Mussatto et al., 2007; Brosig, Whitstone et al., 2007; DeMaso et al., 1991; Sira et al., 2014; Svavarsdottir & McCubbin, 1996; Uzark & Jones, 2003; Visconti et al., 2002); Australia ($N = 5$) (Diffin et al., 2016; Franich-Ray et al., 2013; Hearps et al., 2014; Jordan et al., 2014; Menahem, Poulakis, & Prior, 2008b); Sweden, Netherlands ($N = 3$) (Lawoko & Soares, 2002, 2003b; Levert et al., 2016; Mörelius et al., 2002; Spijkerboer et al., 2007; Utens et al., 2002); Egypt, Korea, Norway, Switzerland ($N = 2$) (Ahn et al., 2014; Arafa et al., 2008; Ezzat et al., 2016; Franck et al., 2010; Goldbeck & Melches, 2005; Grønning-Dale et al., 2012; Grønning-Dale et al., 2013; Helfricht et al., 2008; Lee et al., 2007; Sarajuuri et al., 2012; Werner et al., 2014); Italy, Turkey, England, The United Kingdom, Finland, Saudi Arabia, Germany ($N = 1$) (Almesned et al., 2013; Bevilacqua et al., 2013; Doherty et al., 2009; Yildiz et al., 2009). Twenty-three studies (70%) reported paternal findings with a mean father participation rate of 40% (range 10%-53%). All studies used convenience sampling for parents of a child with CHD with sample sizes ranging from 26–1092. Most studies ($N = 27$, 81%) lacked a theory to guide the research.

Measures

All studies used complete or selected sub-scales of a validated instrument for assessment of parental QOL, with 41 different instruments used overall. Most measures were used in 1–2 studies; however, the General Health Questionnaire (GHQ) with a Cronbach's alpha reliability of 0.94 (Koeter & Ormel, 1991) was used in four studies (Bevilacqua et al., 2013; Menahem et al., 2008a; Spijkerboer et al., 2007; Utens et al., 2002). The full or short version of the Parenting Stress Index (PSI) with Cronbach's alpha

reliability of 0.93 and 0.87 (Abidin, 1995) was used in seven studies (Brosig, Mussatto et al., 2007; Ezzat et al., 2016; Lee et al., 2007; Mörelius et al., 2002; Uzark & Jones, 2003; Visconti et al., 2002).

Parental QOL

Psychological or emotional functioning. Psychological or emotional functioning refers to coping and adapting to a child's health condition and includes the aspects of stress, hope, helplessness, and depression (Varni et al., 2004). Increased stress among parents of a child with CHD was consistently reported in the reviewed studies. Several study results reported increased CHD severity was related to lower psychological scores in stress, anger, depression, fear, hopelessness, and locus of control (DeMaso et al., 1991; Franich-Ray et al., 2013; Grønning-Dale et al., 2013; Hearps et al., 2014; Jordan et al., 2014; Lawoko & Soares, 2002; Menahem, 1998). Parents of a child with CHD reported highest levels of perceived stress when compared to normative scores, parents of children with other health conditions, or parents of healthy children. Generally, CHD mothers demonstrated higher levels of stress than CHD fathers and reported different methods of coping (Ahn et al., 2014; Bevilacqua et al., 2013; Diffin et al., 2016; Doherty et al., 2009; Franck et al., 2010; Franich-Ray et al., 2013; Utens et al., 2000). One study did not observe a statistically significant gender difference in post-traumatic stress disorder (PTSD) scores but demonstrated a decline in PTSD symptoms over time (Helfricht et al., 2008). In fact, numerous studies indicated time to be an important influence in the improvement of psychological factors (Diffin et al., 2016; Franck et al., 2010; Grønning-Dale et al., 2012; Grønning-Dale et al., 2013; Menahem et al., 2008b). One case-control study demonstrated significantly lower total stress scores among CHD mothers compared to mothers of healthy

children and contributed this to the presence of effective adjustment and adaptation (Ezzat et al., 2016). Studies have indicated depression is frequently experienced by CHD fathers, with 20% of CHD fathers reporting depression in comparison to 10% of fathers of healthy children during the perinatal period (Bevilacqua et al., 2013; Giallo et al., 2012; Paulson & Bazemore, 2010).

Physical functioning and well-being. Physical functioning includes the influence a child's health status has on the parent's ability to perform self-care and hygiene, attain adequate, restful sleep, and the experience of headaches, stomach problems, physical pain, and fatigue (Varni et al., 2004). When compared to parents of children with minor illnesses, parents of a child with CHD had lower health scores, role limitations due to poorer physical health, difficulty sleeping, and differences in perceived physical health in maternal and paternal outcomes (Arafa et al., 2008; Bevilacqua et al., 2013; Utens et al., 2000).

Social functioning. Social functioning refers to the ability to maintain relationships both inside and outside of the family unit, including the workplace (Varni et al., 2004). Most studies indicated negative effects on social functioning, although some showed better perceptions of social support by parents of a child with CHD when compared to other parents or at different ages of their affected child (Diffin et al., 2016; Hearps et al., 2014; Lawoko & Soares, 2003b; Lee et al., 2007). Social functioning is influenced by communication and is affected by others not understanding the family's situation, parents' difficulty talking about their child's health condition, having effective conversations with health professionals, as well as a lack of means to allow for communication (no telephone or email access) (Varni et al., 2004). Isolation was found to be a statistically significant correlate among CHD mothers and has been attributed to increased worry about their child's

outcome, side effects, risk of becoming ill, concern about the reactions of others, and the effect the child's condition has on other family members (Sarajuuri et al., 2012; Varni et al., 2004).

Financial stability. Financial stability directly influences access to communication and explained the most variability among social support (9%) in parents of a child with CHD (Lawoko & Soares, 2003b). For example, internet use by CHD mothers for information and social support contributed to lower reports of perceived stress (Lee et al., 2007). A lack of financial stability could mean the inability to afford internet access and would remove a clear source of support for parents of a child with CHD. One study recorded a significant main effect of socioeconomic status on the variance of all scales of the GHQ (Spijkerboer et al., 2007). Nearly all parents of a child with CHD (98%) expressed concern or anxiety about future familial, financial, and health adjustment problems of their children, with 70% complaining of financial problems because of increased expenses related having a disabled child (Almesned et al., 2013; Arafa et al., 2008; Lawoko & Soares, 2002, 2003b; Levert et al., 2016).

Family Functioning

Daily activities. Daily activities address challenges parents experience with providing time-intensive care for their child, completing other household tasks and responsibilities, and attending additional healthcare visits (Varni et al., 2004). Infant feeding was reported to be the most time-consuming task by CHD mothers, with CHD type and long hospital stays contributing to this finding (Svavarsdottir & McCubbin, 1996). CHD fathers designated providing emotional support for the spouse or partner as the most time-consuming and difficult task (Svavarsdottir & McCubbin, 1996). Parents of a child with

CHD reported difficulties maintaining a work-life balance, with increased needs for workplace support and leisure activities (Levert et al., 2016).

Family relationships. The family relationships factor describes problems influencing communication, parent-child interactions, conflicts between family members, parental role fulfillment, and difficulty making decisions and solving problems as a family (Varni et al., 2004). Significant differences in parental role restriction and feelings of decreased competence and relationships were reported during a child's hospital stay and beyond, with CHD mothers expressing significantly higher levels of negative influence than CHD fathers (Brosig, Mussatto et al., 2007; Diffin et al., 2016; Franck et al., 2010; Sarajuuri et al., 2012). Infant attachment and mother-baby bonding did not demonstrate statistically significant differences between CHD mothers and community norms; however, CHD mothers demonstrated increased maternal protectiveness and care (Jordan et al., 2014). Hospital environment, parent age, education levels, underlying genetic defect, lower levels of social support, and long hospital stays were variables associated with reports of increased negative influence on family relationships (Diffin et al., 2016; Hearps et al., 2014; Saied, 2006; Werner et al., 2014).

Gender Differences

There is inconsistency within the literature to sample both parent genders equally and report differences between gender outcomes (Jackson et al., 2015; Lawoko & Soares, 2002; Spijkerboer et al., 2007; Utens et al., 2000). The lack of father participation in one study was speculated to be due to fathers not taking on the burden of their child's surgery to the extent mothers did or fathers not feeling their input was as important or valid since fathers viewed their role to be "busy breadwinners" or their partner's support (Menahem et

al., 2008b). CHD mothers reported lowest availability of social support in contrast to CHD fathers, who reported greatest availability of social interactions (Lawoko & Soares, 2003b). In a conflicting study, CHD fathers reported having to give more things up and see family members and friends less frequently than CHD mothers (Werner et al., 2014). In the majority of reviewed studies, parents of a child with CHD had higher stress levels than control groups; however, CHD mothers tended to have higher stress levels than CHD fathers (Utens et al., 2000). Disengagement was a shared coping strategy for parents of a child with CHD; however, CHD fathers reported higher levels of emotion-focused coping and increased use of alcohol and humor as means of coping (Diffin et al., 2016; Doherty et al., 2009). CHD mothers showed a significantly higher tendency to seek social support as a coping means than CHD fathers (Spijkerboer et al., 2007).

Discussion

This systematic review had two purposes: 1) to identify how parental QOL is affected when having a child with CHD, and 2) to describe factors that influence parental QOL when having a child with CHD. Psychological and emotional experiences such as stress, anxiety, depression, anger, and hopelessness are pervasive among parents of a child with CHD and frequently occur at clinically significant levels or at higher levels than PCOD or PHC (Bevilacqua et al., 2013; Brosig, Mussatto et al., 2007; Brosig, Whitstone et al., 2007; Diffin et al., 2016; Doherty et al., 2009; Ezzat et al., 2016; Franich-Ray et al., 2013; Lawoko & Soares, 2002, 2003b; Sarajuuri et al., 2012; Utens et al., 2000; Uzark & Jones, 2003). An increased sense of isolation and a loss of social support sources among parents of a child with CHD is noted and can be attributed to efforts to care for and protect their child from acquiring illness (Diffin et al., 2016; Doherty et al., 2009; Hearps et al., 2014; Lawoko

& Soares, 2003b; Svavarsdottir & McCubbin, 1996; Visconti et al., 2002; Werner et al., 2014). Parents of a child with CHD report an increase in economic burden due to loss of work or changes in employment to support time needed to care for their medically complex child (Almesned et al., 2013; Arafa et al., 2008; Lawoko & Soares, 2002, 2003b). Parents of a child with CHD report increased financial strain, which influences their perceived QOL and contributes to the level of distress they experience (Almesned et al., 2013; Lawoko & Soares, 2003b). Consequences for the physical health of parents of a child with CHD are demonstrated, with reports of decreased vitality, physical functioning, sleeping challenges, and other physical symptoms (Arafa et al., 2008; Bevilacqua et al., 2013; Sarajuuri et al., 2012; Spijkerboer et al., 2007; Utens et al., 2000). Interactions among family members are reported as being negatively influenced when having a child with CHD (Ahn et al., 2014; Almesned et al., 2013; Brosig, Whitstone et al., 2007; Jordan et al., 2014). Conversely, parents of a child with CHD also report beneficial aspects of having a child with CHD, such as feelings of being closer as a family and having relatives that are more understanding or helpful (Diffin et al., 2016). Parents of a child with CHD also report creative sources for social support and mastery through a variety of coping methods (Diffin et al., 2016; Doherty et al., 2009; Ezzat et al., 2016; Sira et al., 2014; Svavarsdottir & McCubbin, 1996).

The findings of this review are consistent with those in the literature addressing parents of children with chronic illnesses and differences in perceptions and adjustment between mothers and fathers (Hatzmann, Heymans, Ferrer-i-Carbonell, van Praag, & Grootenhuis, 2008; Heaman, 1995; Schilling, Schinke, & Kirkham, 1985). The effect of chronic pediatric health conditions on and treatment of family functioning is a significant concern given the essential role parents and family play in child adaptation (Thompson &

Gustafson, 1996; Varni, Katz, Colegrove Jr., & Dolgin, 1996; Varni & Wallander, 1988). Family strain, parental perceptions, and coping are predictive variables that demonstrate greater influence on a child's adjustment to their illness than the actual illness severity and any physical limitations it causes (Casey et al., 1996; DeMaso, Beardslee, Silbert, & Fyler, 1998; DeMaso et al., 1991). Parents of a child with chronic illness have historically reported lower QOL, and their parent-child interchanges demonstrate a significant effect on family functioning (Cousino & Hazen, 2013; Davis, Brown, Bakeman, & Campbell, 1998; Goldbeck & Melches, 2005; Lawoko & Soares, 2003a; Mussatto, 2006; Streisand et al., 2001). This review demonstrates that parents of a child with CHD frequently report even greater QOL effects in comparison to parents of children with other chronic illnesses. This is significant, as parental mental health has been found to moderate the physical and psychosocial aspects of QOL in their child with CHD and is consistently related to the increased risk of child maltreatment and developmental differences in the child (Dulfer et al., 2015; Kennedy, 2012; Levert et al., 2016). A study looking at child behavior outcomes found that parents of a child with CHD with bonadaptation and effective coping perceived their children with CHD to have "significantly fewer internalizing problems" (Visconti et al., 2002).

Assessment of needs or changes of parental QOL can be achieved through early and periodic evaluation using a comprehensive instrument. This review points out the number of instruments available that measure only selective QOL factors. The selective characteristic of QOL instruments, coupled with research that lacks cohort or interventional designs, contributes to a gap in understanding efficacious treatments and QOL changes over time in this patient population. Strength of evidence presented is limited on the level of single-site

studies with small sample sizes and lack of gender or racial diversity but is relatively representative of the research that has been performed on parents of children with other chronic illnesses (McCubbin & McCubbin, 1996).

There is an array of affected QOL factors with varied intensities among both genders of parents of a child with CHD. Parents of a child with CHD generally report higher levels of stress that affect one or more QOL factors that indirectly influence other QOL factors. Gender appears to influence which QOL factors are affected based on the roles performed or perceived role expectations held by the parent. Understanding gender differences in parental perceptions of QOL has long been a research concern, with goals to promote intervention placement that improves stress management, coping, and family adjustment for mothers and fathers (Beckman, 1991; Gray, 2003; Heaman, 1995; Perry, 2004; Trute, 1995). Most cardiac-related pediatric outcomes literature that includes parental measures emphasizes the maternal responses to or the effect the child's care or condition has on mothers, often paying little attention to the influence of, or effect on, the father (Engle et al., 2011; Schilling et al., 1985). This informational gender gap can contribute to difficulties for health care providers to identify a parent's unique area of need for supportive interventions. Economic influences, cultural shifts in social media, television, and internet usage by fathers for support and education are indicators that fathers are more involved with their child's care than ever before and need to be represented within the research (Coleman, Garfield, & Committee on Psychosocial Aspects of Child and Family Health, 2004; Raeburn, 2014).

Father involvement with their child is important for the child's overall development and wellbeing. Children with chronic illnesses, such as CHD, affect fathers uniquely, and this population is vastly under-represented in research and thereby lacking in equally

representative policies. Development of father-inclusive policies that are supported by research and reflective of the culture shifts and economic trends can go far in the acceptance and promotion of the new, nurturing, co-parenting father (Sarkadi et al., 2008). Historically, recruiting fathers to undergo treatment for psychological distress has been difficult, and engaging them in treatment can be even more challenging as men can be unwilling to seek help for mental health concerns, express negative attitudes about therapeutic interventions, are less likely to make doctor visits, and are more likely to discontinue therapy compared to women (Addis & Mahalik, 2003; Mansfield, Addis, & Mahalik, 2003; O'Brien et al., 2016; Primack, Addis, Syzdek, & Miller, 2010). Although participation in interactive relationships may be difficult for some fathers to adopt as women traditionally seek social support through relationships, it should be encouraged as it promotes effective personal coping and improves their child's overall outcomes (Bruce et al., 2016; Connell, 2005).

Limitations among reviewed studies include the presence of biases or the lack of bias discussion, small sample sizes, and the absence of theory use for structural framework and discussion of sample size determination methodology (i.e., power analyses). The following theories were used among six of the reviewed articles to provide a structural framework and guide variable selection and data analysis when there were quantitative findings (Ezzat et al., 2016): the double ABCX model of family adjustment and adaptation (McCubbin & Patterson, 1983b), the transactional stress and coping model (Lazarus & Folkman, 1984), the resiliency model of family stress, adjustment, and adaptation (McCubbin & Patterson, 1983b) , and parenting stress index model (Abidin, 1995).

Nursing Implications and Conclusions

This systematic review punctuates the need for a multi-factorial approach to improve parents of a child with CHD QOL. Nursing research using theory as a framework to guide the research can add value to the data, guide practice, and promote outcomes through identification and explanation of QOL phenomena in CHD families (Polit & Beck, 2008). Nursing practice implications include use of family-centered care with multidisciplinary collaborations to acknowledge the needs of each parents of a child with CHD. Clinical identification of affected parents of a child with CHD QOL factors can enhance timely, appropriate, and supportive interventions to improve coping and adaptive efforts (Chock & Lee, 2014; Soulvie, Desai, White, & Sullivan, 2012). Longitudinal studies should also be considered to further describe correlations of parents of a child with CHD QOL with their child's developmental outcomes. Continued research efforts using father-inclusive/specific care models will explain and identify unique QOL experiences and assist in recognizing and encouraging fathers' help-seeking behaviors and support changes in policy to support fathers' needs (O'Brien et al., 2016). Promotion of parents of a child with CHD QOL is an earnest endeavor with great potential to improve the well-being and developmental outcomes of all family members.

CHAPTER 3

THEORETICAL FRAMEWORK AND METHODOLOGY

This chapter describes McCubbin and McCubbin's (1983a) double ABCX model of family stress and adaptation, the theoretical framework, and how it was used as the foundation of this study. The evolution of the ABCX model from Hill's (1949) family stress theory and components of the model and its application to CHD families are also discussed. The methodology used in the PinCHeD study concludes the chapter.

Theoretical Underpinnings

Application of theoretical frameworks or models is used to assist in research design, guide variable selection, and provide structure to quantitative data analysis (Ezzat et al., 2016). Mussatto's (2006) work provides a review of theoretical approaches used to study family adaption when having a child with a chronic illness. - The review focuses on the adaptation-related experiences of parents of a child with CHD and concludes with families of a child with CHD who experienced numerous and diverse stressors and coping aspects that should all be taken into consideration when assessing adaptation and determination of most helpful interventions. Over the years, various theories, conceptual models, and grounded theory approaches have been used or developed to assist in the understanding of the challenges and perspectives experienced by the families of children with CHD in their development of resilience and bonadaptation (Lisanti et al., 2018; Rempel et al., 2012).

One such model is Lisanti's (2018) individualized family centered developmental care model for application to the period when a child with CHD is receiving care in the pediatric cardiac intensive care unit. Lisanti et al.'s (2018) individualized family-centered developmental care model was developed for application in the pediatric cardiac intensive

care unit to promote the infant's development during this time of hospitalization. Lisanti et al. (2018) outline the CHD-specific risk factors for the infant and appropriate family-centered interventions to be considered to support the development of the infant. Although not specifically for understanding the needs of parents, many of the family-centered interventions suggested by the individualized family centered developmental care model are useful in addressing the psychological and support needs of parents (Lisanti et al., 2018).

Another model is Rempel et al.'s (2012) constructivist "parent under pressure" grounded theory, which addresses the parenting processes when having a child with hypoplastic left heart syndrome (HLHS) during the period of time from diagnosis until the child undergoes the second palliative surgery. Rempel et al.'s (2012) grounded theory was conducted to generate a model to better understand the unique experiences of these parents and to improve clinical management of their needs. A strength of Rempel et al.'s (2012) work was the inclusion of grandparents of the child with HLHS, as they frequently serve as a support source to the parents. Both the family centered developmental care model (Lisanti et al, 2018) and the parenting under pressure grounded theory (Rempel et al., 2012) are excellent options for guiding practice and supports when working with parents and families of a child with a specific CHD or the hospitalized child which limits their scope of application and are not appropriate choices for the long term perspective that was needed in the PinCHed study.

The double ABCX theory of family adjustment and adaptation (McCubbin & Patterson, 1983a, 1983b) was selected to guide the PinCHeD study. Unlike Lisanti et al's (2018) model, which is to be applied during the child's time in the pediatric cardiac intensive care unit, or Rempel's (2012) grounded theory, which is specific to parents of a

child with HLHS, the double ABCX theory of family adjustment and adaptation (McCubbin & Patterson, 1983a, 1983b) can be applied to the family over the life course of the child with any CHD type. The double ABCX model (McCubbin & Patterson, 1983a, 1983b) accounts for changes over time for the family of a child with CHD, provides a platform for continuous monitoring of family adjustment, guides identification of needs among family members, and outlines the relationships among the family members' perceptions of the stressor and supports in place. Finally, the double ABCX theory (McCubbin & Patterson, 1983a, 1983b) includes all factors related to stress and adaption: the stress-producing stimuli, the family's resources, perceptions of the stressor and resources, the crisis, coping, and overall adaptation for families of a child with CHD and is applicable to longitudinal studies when assessing family adaptation over time.

Theoretical Framework

McCubbin and Patterson's (1983a, 1983b) double ABCX theory of family adjustment and adaptation was chosen as the framework for this study. The double ABCX theory describes the role of nursing as one that promotes family members' health, recovery from illness, and maximum functioning within specific health limitations (McCubbin & McCubbin, 1993). The double ABCX theory leads to interventions that support and enhance family strengths, assists families in maintaining connections with community supports, and aids families in arriving at a realistic appraisal of what is the best fit for them in their situation (McCubbin & McCubbin, 1993).

Theoretical Evolution

Family resilience has been a focus of crisis and stress research since the 1920s, but it was Hill's (1949) family stress theory and associated ABCX model of family stress that

served as the reference point for most of the subsequent family resilience theory developments (Nichols, 2013). The family stress theory was developed by Hill (1949) as a result of his work in explaining "family dismemberment" during and after World War II. Hill, a social scientist working for the Army, was charged with assessing the impact of war casualties on American families. Hill's (1949) ABCX theory of family stress is a middle-range theory, and, though modified, is still used in family development to describe the process by which families adapt and endure through crisis over the lifespan. One of the concerns about this theory was the difficulty this linear approach had in explaining complex families and stressors—it addressed only one stressor and its impact and course on one family at a given time. Families typically experience multiple stressors over time. One of the strengths of this theory was that it is applicable to real-life situations and can be a helpful tool in developing effective therapies for families.

Due to these limitations, McCubbin and Patterson (1983a) adapted the Hill family stress model to become the double ABCX model, making it more dynamic in order to address concepts such as crisis and adaptation over time. Their model, the double ABCX model, added a post-crisis stage to illustrate the constant adaptation experienced by families during and in the period of time surrounding crisis. McCubbin and McCubbin (1993) expanded this model to include five propositions that described relationships found within the model itself (Freidman, 1998; McCubbin & McCubbin, 1993). The five propositions are: a) there is a positive relationship between the severity of strain associated with a crisis that is influenced by other stressors and strains on the family; b) there is a negative relationship between the level of adaptation of the family to the crisis and the influence of the severity of the pileup of stressors and strains; c) there is a positive relationship among the amount of

personal and family system resources, and social support the family has, and the influence those variables have on the adaption to the pile-up of stressors and strains; d) there is a negative relationship between the amount of personal and family system resources and social support influences, and the severity of strain created by the pile-up of demands, and e) a positive relationship exists between the level of the family's sense of coherence regarding the total situation and the family's adaptation (Lavee, McCubbin, & Patterson, 1985).

The double ABCX model (McCubbin & Patterson, 1983b) illustrates the constant adaptation experienced by families in crisis and is a pre-cursor to the resiliency model of family adaptation and adjustment (McCubbin & McCubbin, 1993). The double ABCX model can be used to explore causal relationships among stressors, perceptions, resources, and adaption (Lavee et al., 1985). Middle-ranged theories, like the double ABCX theory, work best in framing studies because they have the advantage of being narrow in scope and have testability among the concepts (McEwen, 2014). An advantage of using the double ABCX model is its ability to provide a framework to describe the relationships of social support and other family assistive resources while explaining the family's level of coping and adaptation at a given point over time. The double ABCX model (McCubbin & Patterson, 1983a) also describes the phenomenon of a "pile-up" of stressors and could be used during longitudinal studies.

Role of Nursing

McCubbin and McCubbin describe the role of nursing within the family stress theory as promoting family members' health and recovery from illness, maximizing functioning within specific health limitations, supporting and enhancing family strengths, assisting

families in maintaining connections with community supports, and aiding families in arriving at a realistic appraisal of what is the best "fit" for them in their particular situation (McCubbin & McCubbin, 1993).

Theory Factors

Hill's (1949) associated ABCX model of family stress depicts the dynamics in families that were key elements to adjustment and maladjustment during war, separation, and reunion. Hill's research discovered that it was the interface between stressors (A), resources (B), and the perception of the event (C) that determined how the crisis (X) was experienced and managed in any given family (Joseph, Goodfellow, & Simko, 2014). McCubbin and Patterson (1983a) built upon Hill's ABCX model by adding a post-crisis concept in their double ABCX model. In the double ABCX model, Hill's original factors of ABCX are represented in lowercase, as those original factors are still present post-crisis in addition to the new, capitalized, post-crisis factors determined by McCubbin and Patterson. Figure 2 depicts the double ABCX model when applied to families of a child with CHD. The model works from left to right, demonstrating the passage of time. Descriptions and examples of each part of the figure are described next.

A and aA Factors

A and aA factors (see Figure 2) are the stimuli that produce the stress response in a parent or family. Examples of A factors include sharp changes in income, illness of a family member, child-discipline challenges, a change in the roles of the parents (a wife taking on both parental roles due to husband's absence), and changes in living situations. aA factors represent a "pile up" of stressors and describes hardships or complications that persist to become chronic conditions (Weber, 2011). Examples of aA factors for this population

include disapproval from other family members regarding the decisions made by parents pre-crisis or as a result of the crisis, child behavior problems, and role conflict parents may face when they have one child in the hospital for an extended period of time while other children remain at home (Baker, Blachar, Crnic, & Eldelbrock, 2002).

B and bB Factors

Hill (1958) referred to B and bB factors (see Figure 2) as *the family's stressor-meeting resources* and their presence or absence, which either kept the family from reaching crisis or urged them into crisis. Examples of B factors include self-reliance, social support, financial adequacy, friends, religious activities, and family strength. One B factor that is often experienced by families with a child who has CHD is social isolation, as parents may shield their child from being in contact with contagion carried by others that could seriously impact their child's health. Resource factors that existed prior to the initial crisis (b) and new resources (B) that developed while the family was coping with the initial crisis are labeled as 'bB' (Weber, 2011). Examples of new resources include improved self-esteem, reallocation of roles and responsibilities, new treatment by therapeutic or mental health professionals, increased flexibility, practice of new traditions and celebrations, increased power of endurance, and spirituality (Joseph et al., 2014; Weber, 2011).

C and cC Factors

C and cC factors (see Figure 2) are the family's perceptions of the stressor (A or aA) and their ability to meet the resources (B or bB) required to prevent a crisis from occurring or after the initial crisis has occurred (Hill, 1958; McCubbin & Patterson, 2007). Examples of C factors include parent efficacy, family strength, and cohesiveness. C factors have been described as meaning-making factors in that they resemble how a family makes sense of the

stress they are experiencing (Darling, Senatore, & Strachan, 2012). Whereas one family may view having a child with CHD as an insurmountable burden, another may view the challenge as a "blessing in disguise." cC factors are the perceptions of all preceding factors combined and may be very different than the perceptions parents held prior to the initial crisis. These new perceptions, if positive, may reflect new growth, improved ability to overcome challenges, and improved family unification. If negative, they may reflect parental feelings of defeat, inadequacy, and lack of competence. This study provides information relative to C and cC factors (circled in Figure 2).

X Factor

The X factor (see Figure 2) is a representation of the experienced crisis. Crisis has been operationally defined as disruption or breakdown in a person's or family's normal or usual pattern of functioning that cannot be resolved using a person's customary problem-solving resources or skills (Washington State Department of Social and Health Services, 2019). Examples of X factors when applied to families of a child with CHD include the moment parents receive their child's diagnosis of CHD or when the child undergoes surgery to palliate their CHD.

Coping

Coping (see Figure 2) is the practice or skill of using both cognitive and behavioral strategies to facilitate and navigate the post-crisis. Coping efforts may be directed at (a) elimination or avoidance of stressors, (b) management of the situation's hardships, (c) maintenance of the family system's morale and integrity, (d) acquisition or development of resources to meet needs, and (e) implementation of changes in the family system to meet the new needs (McCubbin, 1979; McCubbin & Patterson, 1982).

xX Factor

The xX factor (see Figure 2) represents overall adaption and exists on a continuum. Normal function indicates bonadaptation (Joseph, 1989), while examples of maladaptation for this population include problems with family relationships, such as poor communication and conflicts between family members, and difficulty making decisions and solving problems as a family.

The double ABCX theory of family adjustment and adaption (McCubbin & Patterson, 1983a, 1983b) was integrated into the PinCHeD study's research design through the selection of the instruments used to measure parental perceptions of their stress, QOL, and family functioning as well as the statistical analyses used to examine the strength of relationships between family functioning and parental stress and QOL. The methodology used in the PinCHeD study is described in the ensuing section.

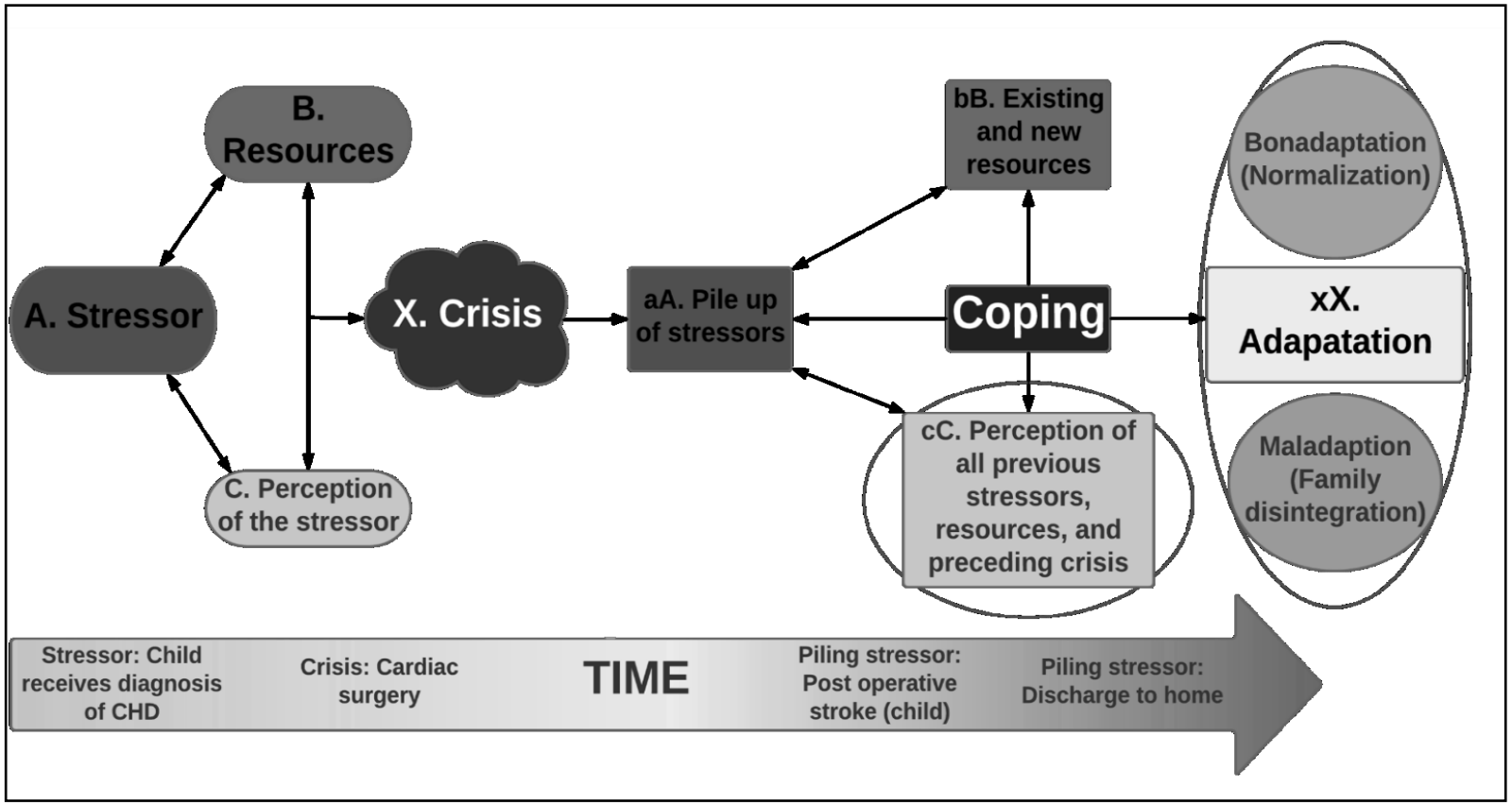


Figure 2. Application of the double ABCX model of family stress and adaptation to families of a child with CHD (Adapted from McCubbin and McCubbin's [1983] double ABCX model).

Methodology

Design

A descriptive, correlational, cross-sectional design was used in The PinCHeD study to determine how mothers and fathers of a child with CHD perceive their personal levels of stress, QOL, and family functioning, how these levels differ between parent pairs, how stress and QOL is related to family functioning, and how severity of infant CHD is related to parental stress, QOL, and family functioning.

Setting

The setting for the PinCHeD study was Children's Mercy Hospital (CMH) in Kansas City, Missouri. Children's Mercy Hospital is a free-standing, 355-bed academic pediatric medical center located in Kansas City, Missouri, that provides comprehensive primary and tertiary specialty care to children in Missouri and Kansas. It is the only pediatric medical center between St. Louis and Denver and provides comprehensive care in 50 pediatric subspecialties. The hospital is the only Level I pediatric trauma center in the region and is the primary pediatric teaching hospital for the University of Missouri-Kansas City (UMKC) School of Medicine. The Ward Family Heart Center at CMH provides comprehensive, state-of-the-art inpatient/outpatient care and research for cardiovascular disease. U.S. News and World Report (2018) ranked pediatric cardiology at CMH as the 19th best program in the nation. Over 400 operations are performed annually, ranking CMH as a high-volume cardiothoracic surgery center for children, with publicly reported outcomes that exceed the national benchmark (Society of Thoracic Surgeons, 2018).

The Cardiac Neurodevelopmental (CND) Program at CMH was started in 2013 with a purpose of monitoring children with cardiac conditions for their neurodevelopmental

outcomes. The CND program is an interdisciplinary program of collaborating providers spanning developmental and behavioral, neurology, cardiology, hearing and speech, occupational and physical therapy, and social work disciplines. This study served as the program's initial research project and a pilot study to test feasibility of the research, accessibility to the parents of children who receive neurodevelopmental assessments at CMH, and the capability and readiness of the Heart Center data repository to support larger longitudinal studies that measure parent and child neurodevelopmental outcomes.

Sample

After hospital IRB approval was granted, a purposive sample of 62 parents whose child with CHD received neurodevelopmental evaluations under the age of six from CMH's CND program were recruited by the primary investigator (PI) during the study period of June 1, 2013 through December 31, 2017. The nurse coordinator of the CND program was the PI for this study and had full access to this patient population (see Appendix B for letters of support). As suggested by the hospital IRB, the participants in this study were separated into primary (parents) and secondary (children) samples.

Primary population-inclusion criteria.

1) Parent pairs of a child with CHD who has received a neurodevelopmental evaluation at or below six years of age by CND program providers between the dates of June 1, 2013 and December 31, 2017. The term "parent" refers to biological or adoptive parents, step-parents, or other legal guardians with whom the child with CHD resides. Pregnant mothers were included.

a. Parent pairs are two people who identify themselves as a couple, are married and/or reside together. For the purpose of this study, the legal definition of "marriage" was used:

    i. The legal union of a couple as spouses. The basic elements of a marriage are: (1) the parties' legal ability to marry each other, (2) mutual consent of the parties, and (3) a marriage contract as required by law (Marriage, 2017).

2) The child's family must have resided within the hospital catchment area (mostly Kansas and Eastern Missouri).
3) Parents who speak and understand English or Spanish (as noted during initial CND program intake process).

Primary population-exclusion criteria.

1) Parents of a child with CHD who received a neurodevelopmental evaluation at or older than six years by CND program providers between the dates of June 1, 2013 and December 31, 2017
2) Parent pairs that do not reside together
3) The child's family relocated outside of the hospital catchment area
4) Adults are unable to consent
5) Individuals who are not yet adults (infants, children, teenagers)—primary population only
6) Prisoners
7) Wards of the state

Secondary population-inclusion criteria.

1) A child with CHD of parental pairs who participated in Phase 1 who has received a neurodevelopmental evaluation at or below six years of age by CND program providers between the dates of June 1, 2013 and December 31, 2017.

Secondary population-exclusion criteria.

1) Any child with CHD who received a neurodevelopmental evaluation, by CND program providers between the dates of June 1, 2013 and December 31, 2017 whose parents (both) did not participate in Phase 1.

An a priori power analysis for a matched paired t-test using G Power software (Faul, Erdfelder, Lang, & Buchner, 2007) was performed to assist in determining sample size needed to detect a statistically significant difference between outcomes of mothers and fathers in this study. Using an alpha of 0.05 and an effect size of 0.50 (considered a "medium" effect), a total sample size of 34 parent pairs ($N = 68$) was needed for a power of 0.80. A power of 0.80 indicates an 80% chance of rejecting the null hypothesis (Cohen, 1977).

Instruments

Demographic Survey

Using the parent demographic survey, the PI gathered the following information regarding parents of a child with CHD: highest education level obtained, annual household income, gender identification, sexual orientation, birth date, relationship status and duration, race and ethnicity, parent type (biological, step/bonus, adoptive, legal guardian/foster parent). Although several research questions within this study were binary, attention was paid in the design of the demographic survey tool to ensure no parent/caregiver meeting

inclusion criteria was unintentionally excluded or inaccurately represented in data collection based upon sexual orientation or gender identity. Benchmark resources from the Fenway Institute (2018) were consulted in the survey development regarding sexual orientation and gender identification questions to allow for parents of non-binary families to indicate their gender identification and sexual orientation. Household types and relationships within the demographic survey used the definitions and categories found within the most recent United States Census Bureau coding documents (United States Census Bureau, 2016, 2017).

Child Demographics

Child demographics were collected from the Heart Center data repository (IRB#13020045) or through chart review after their parents' responses were received. Data related to the child's medical condition such as fundamental cardiac diagnosis, non-cardiac anatomic abnormalities, chromosomal abnormalities, syndromes, neurodevelopmental test scores, whether or not the child had had abnormal brain imaging findings or received early intervention services, highest in-hospital mortality risk measure associated with cardiac surgery completed between birth and date of neurodevelopmental testing, most recent open heart cardiac surgery, gender, and child's age. The child's fundamental cardiac diagnosis, chromosomal abnormalities (if any), and syndromes (if any) were coded and described as indicated by the Society of Thoracic Surgeons database, version 3.3 (2015).

Parenting Stress Survey

The Pediatric Inventory for Parents (PIP) (Streisand et al., 2001) was used to measure parenting stress. The PIP was developed by pediatric psychologists with the intention to measure stress in parents of children experiencing any chronic illness (Streisand et al., 2001). Parents rated the perceived frequency (PIP-F) and difficulty (PIP-D) of 42

stressful events associated with parenting a child with a chronic illness using two, 5-point Likert-type scales ranging from "never" to "very often" (frequency) and "not at all" to "extremely" (difficulty). Parenting stress was measured across four scales: communication, medical care, role functioning and emotional distress (Streisand et al., 2001). Each scale's possible scores ranged from 42 to 210, with higher scores indicating greater perceived parenting stress related to more frequent and more difficult stressors, respectively (Hilliard, Monaghan, Cogen, & Streisand, 2011). The PIP has demonstrated adequate validity in frequency and difficulty (r^2 = 0.43 and 0.45 respectively) and reliability (α = 0.80-0.96) in parent populations of healthy and chronically ill children for both scales (Streisand et al., 2001). The PIP is a well-established measure that has been used without modifications in parents of children with diverse pediatric chronic health conditions, including pediatric cardiology (Alderfer et al., 2008; Hilliard et al., 2011; Kaugars et al., 2018; Vrijmoet-Wiersma et al., 2009).

Quality of Life and Family Functioning Survey

The Pediatric Quality of Life Inventory™ Family Impact Module (PedsQL™ FIM) is a QOL and family functioning questionnaire for parents of children with chronic conditions (Knez, Stevanovic, & Vulić-Prtorić, 2017; Varni et al., 2004). The PedsQL™ FIM is a self-rated, 36-item questionnaire with eight scales: physical functioning (6 items), emotional functioning (5 items), social functioning (4 items), cognitive functioning (5 items), communication (3 items), worry (5 items), daily activities (3 items) and family relationships (5 items) (Varni et al., 2004). All items use a 5-point Likert-type scale (ranging from 0 = never a problem to 4 = almost always a problem), all are reverse-scored, and linearly transformed to a 0–100 scale so higher scores indicate better QOL and family

functioning. Both instruments exceeded minimum reliability standard of 0.70, and most approached or exceeded the reliability criterion of 0.90 recommended for analysis of individual patient scale scores (Pedhazur & Schmelkin, 2013) (see Table 3).

Instrument Integration with Double ABCX Model

Instrumentation used to measure parental perspectives for the PinCHeD study was guided by the theoretical framework. The double ABCX model captures characteristics of stress, QOL, and family function within several of the factors in the model and illustrates the relationships between these factors in their response to A (Stressor) or aA (pile up of stressors). The subscales and summary scores measured by the PIP are represented as aspects of factors C (perception of the stressor) and cC (perceptions of all previous stressors, resources, and preceding crisis) within the double ABCX model of family adaptation and adjustment (McCubbin & Patterson, 1983a, 1983b). Additionally, factors C and cC are also represented by the worry, daily activities and family relationships subscales of the PedsQL FIM. Parental perceptions of their QOL, measured in the PedsQL-FIM physical functioning, emotional functioning, social functioning, cognitive functioning, and communication subscales and HRQOL summary score, are represented in factors B (resources) and bB (existing and new resources) in the model as they include aspects of support, self-reliance, financial adequacy, and religious activities. The overall results of the parent reports are represented in the xX factors as they describe the current level of adaptation and adjustment the parent endorses for their situation with low levels of stress, high QOL, and high family functioning scores indicating bonadaptation (xX factor).

Table 3 delineates the reliability and validity scores of the English and Spanish versions of instruments used in the PinCHeD study. Spanish versions of all surveys were

available to accommodate eligible families who were primarily Spanish-speaking. Completion of the instruments by each parent took about 30 minutes.

Table 3

Reliability and Validity Scores of Instruments Used

Measure	Reliability	Validity
PIP-English -Stress	α = 0.80-0.96 (Streisand et al., 2001)	Frequency (r^2 = 0.43) Difficulty (r^2 = 0.45) (Streisand et al., 2001)
PIP-Spanish -Stress	α = 0.92-0.94 (del Rincón, Remor, & Arranz, 2007)	Frequency (r^2 = 0.77, p = 0.00) Difficulty (r^2= 0.77, p = 0.00) (del Rincón, Remor, & Arranz, 2007)
PedsQL-FIM-English -Quality of Life -Family Function	α = 0.97 (Total scale score); α = 0.82-0.97 (subscale score range) (Varni et al., 2004)	Construct validity effect sizes 1.08 (Total scale score) 0.19- 1.45 (subscale score range) (Varni et al., 2004)
PedsQL-FIM-Spanish -Quality of Life -Family Function	α = 0.97 (Total scale score); α = 0.82-0.97 (subscale score range) (Scarpelli et al., 2008; Varni et al., 2004)	Construct validity effect sizes 1.08 (Total scale score) 0.19- 1.45 (subscale score range) (Varni et al., 2004)
Demographics Survey	N/A	N/A

Procedures

Letters of support were received from the Cardiac Neurodevelopmental Program Research Director and Director of the Ward Family Heart Center. Institutional Review Board (IRB) approval (#STUDY00000219) was obtained from CMH along with an agreement to rely on a partner institution from the University of Missouri-Kansas City. Eligible families were identified by the PI, and parents were mailed study informational letters with postage-paid response cards to remit to the PI indicating their interest in participating. Contact

information of the PI was provided to parents seeking additional information about the study. Information contained within these letters included the purpose of the research study, data to be collected, and approximately how long the surveys would take to complete. Parents who indicated interest in participating were mailed completion instructions, the demographic survey, instruments, and a postage-paid, pre-addressed envelope (PPE) for returning completed forms to the PI. To mitigate attrition effects, reminder letters were sent by the PI every 30 to 45 days with a maximum of two reminders to parents who had not yet returned completed surveys.

Data Analysis

All completed instruments were scored by the PI. Instrument responses and demographic information were entered into the neurodevelopment section of the secure Heart Center repository for storage by the PI. Heart Center is a web-based application and repository that is accessible only on the CMH network that uses standard secure socket layer (SSL)-certificate secured communication between client and server. All data extractions and transfers from the Heart Center repository to IBM SPSS version 24 took place on the CMH network behind the firewall. Data containing patient identifiers remained on CMH approved and protected devices. Security policies prevent the storage or transfer of data to non-encrypted removable media.

Data were extracted and exported into SPSS and cleaned by the PI with assistance from a UMKC School of Medicine biostatistician. The PI performed the statistical analyses with oversight by the biostatistician. Descriptive statistics were generated to describe general characteristics of the parents (primary sample) and children (secondary sample). Alpha level of 0.05 was used for statistical significance for all analyses unless stated

otherwise. When appropriate, effect sizes were determined to illustrate meaningful (clinical) significance. Due to the small sample size and non-normative distribution, non-parametric statistics were used when testing the proposed hypotheses. Table 4 outlines each hypothesis, the independent and dependent variables, and statistical tests used.

Table 4

Variables and Statistical Tests for Analysis of Hypotheses

Hypothesis	Variables	Statistical test(s) Used
(H1a) Mothers of a child with CHD will report higher levels of stress compared to fathers. (H1b) Mothers of a child with CHD will report poorer QOL compared to fathers. (H1c) Mothers of a child with CHD will report lower levels of family functioning compared to fathers.	Independent- • Parent Gender (Categorical) Dependent- • PIP responses (all subscales and summary scores) (Ordinal) • PedsQL-FIM responses (all subscales and summary scales) (Ordinal)	Wilcoxon Matched Pairs
(H2a) Parents who report high levels of stress will also report low family functioning levels. (H2b) Parents who report poor QOL will also report low family functioning levels.	Independent- • Parent responses PIP (all subscales and summary scales) (Ordinal) • PedsQL-FIM (QOL scales & summary scales) (Ordinal) Dependent- • PedsQL-FIM Family functioning summary scale (Ordinal)	Spearman Correlation
(H3a) Parental stress levels will be higher when their child has a more severe type of CHD. (H3b) Parents will report poorer QOL when their child has a more severe type of CHD. (H3c) Parents will report lower family functioning levels when their child has a more severe type of CHD.	Independent- • STAT score (Ordinal) Dependent- • PIP responses (all (all subscales and summary scales) (Ordinal) • PedsQL-FIM responses (all subscales and summary scales)) (Ordinal)	Spearman Correlation

This chapter reviewed other theories that were considered for this study and provided rationale for selecting McCubbin and Patterson's (1983a, 1983b) double ABCX theory of family adaptation and adjustment. A description of McCubbin and Patterson's (1983a, 1983b) double ABCX theoretical framework was provided to explain its application as the foundation of the PinCHeD study. Double ABCX factors were outlined to assist with how instrument selection was made for surveying parental perceptions when having a child with CHD. The methodology used in the PinCHeD study concludes the chapter. The next chapter discusses the participation response rates, sample demographics, preliminary analyses for assumption testing, and findings for each proposed hypothesis and additional analyses performed.

CHAPTER 4

RESULTS

Chapter 4 provides a report of the findings related to the research questions. This chapter is organized by a discussion of participation response rates, sample demographics, preliminary analyses for assumption testing, and a summary of the findings for each proposed hypothesis.

Participation Rates

Participants were recruited from November 26, 2018 to March 22, 2019. Two hundred eighteen families were identified as meeting eligibility criteria and were sent recruitment letters. Eligibility recruitment and response rates are illustrated in Figure 3. The sample consisted of 31 parent pairs (n = 62) and their child with CHD (n = 31).

Child Demographics

The children with CHD of the participating parents were mostly male (n = 19, 61%). The child's mean age at time of parent survey completion was 4.83 years. The mean difference between the time of parent survey completion and their child's first cardiac surgery was 4.46 years, and the time between the survey and their child's most recent surgery was 3.43 years. The mean number of cardiac surgeries each child has experienced was 2.03 with a range of 1-4. There were a total of 18 unique fundamental cardiac diagnoses with a range that spanned all levels of severity (1-5). Severity of CHD score distribution was slightly skewed to the left (-0.663), indicating more children had CHD types of higher morbidity and greater complexity (Jacobs et al., 2012; O'Brien et al., 2009).

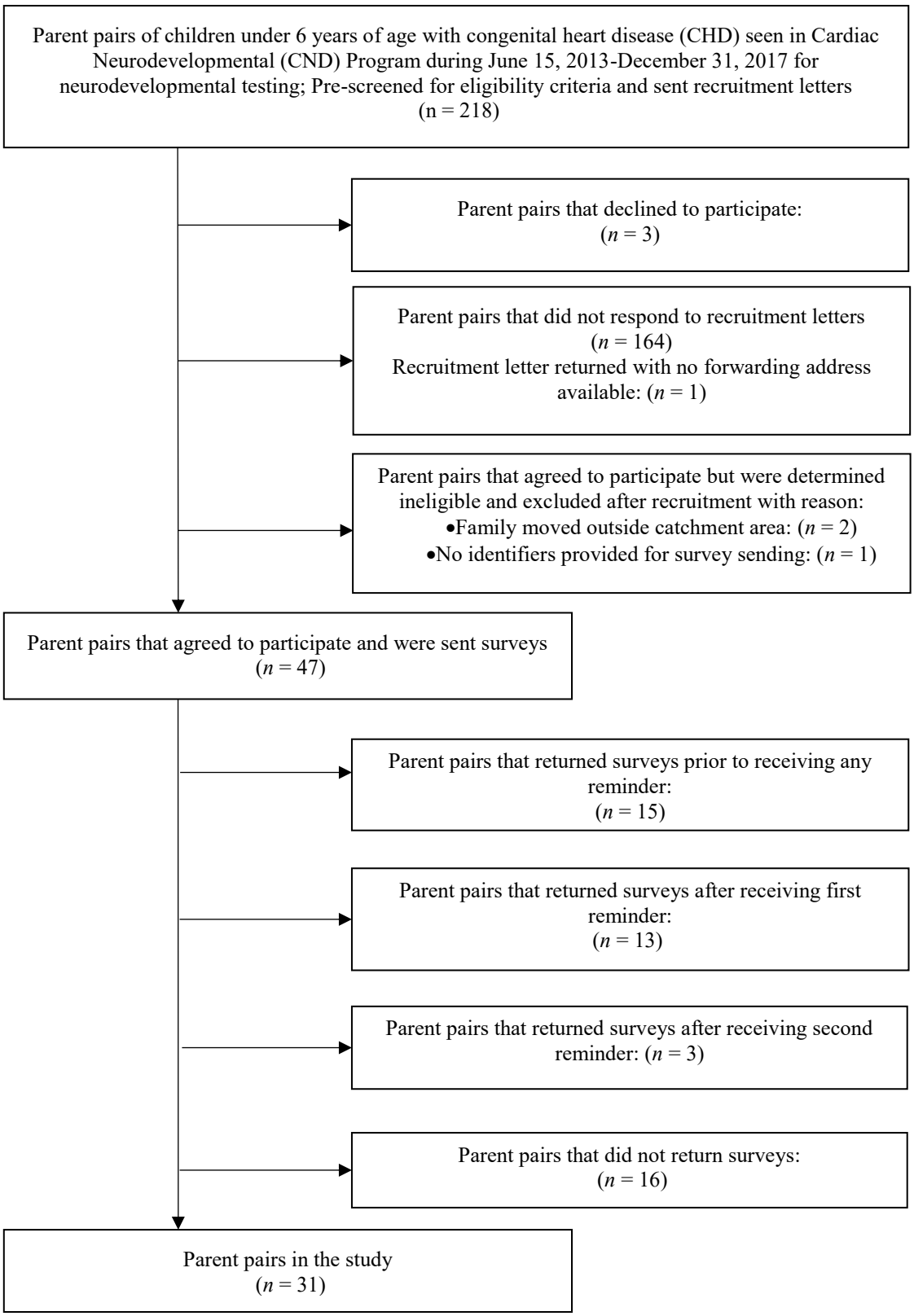


Figure 3. Eligibility and recruitment flowchart.

The average times elapsed from first and most recent surgeries were approximately 4 ½ years and 3 ½ years, respectively. The majority of children with CHD had undergone two or fewer surgeries requiring cardiopulmonary bypass (*n* = 21, 67.7%). Over half (*n* = 18, 58.1%) had STAT scores four or five, indicating diagnosis of CHD type with high mortality and morbidity risk. Of these children, (*n* = 19, 61.3%) did not have chromosomal abnormalities and 22 (71%) did not have a history of abnormal brain imaging. The majority of children with CHD (*n* = 23, 74.2%) received early intervention services, indicating presence of an early developmental delay or difference. Child demographics are provided in Table 5.

Table 5

Child Demographics

Characteristics	Mean ± SD or Frequency (%)
Gender (Male: Female)	19 (61.3%): 12 (38.7%)
Age (years)	4.83 ± 2.03
Time since 1st CV surgery (Years)	4.46 ±2.18
Time since most recent CV surgery (Years)	3.43 ±2.28
Total number of CV surgeries	
1	13 (41.9%)
2	8 (25.8%)
3	6 (19.4%)
4	4 (12.9%)
Early Intervention program participation (No: Yes)	8 (25.8%): 23 (74.2%)
Highest STAT score	
1	6 (19.4%)
2	1 (3.2%)
3	6 (19.4%)
4	10 (32.3%)
5	8 (25.8%)
Fundamental CHD Diagnosis (by category)	
Septal Defects	7 (22.6%)
Pulmonary Venous Anomalies	2 (6.5%)
Right Heart Lesions	6 (19.4%)
Left Heart Lesions	5 (16.1%)
Single Ventricle	2 (6.5%)
DORV	5 (16.1%)
DOLV	1 (3.2%)
Thoracic Arteries and Veins	3 (9.7%)
Chromosomal abnormalities (No: Yes)	19 (61.3%): 12 (38.7)
Syndromic (No: Yes)	27 (87.1%): 4 (12.9%)
History of Abnormal Brain Imaging (No: Yes)	22 (71%): 9 (29%)

The fundamental child cardiac defect types were diverse, with only 25.8% (n = 8) being defects with single ventricle pathophysiology. A complete breakdown of fundamental CHD types and frequencies among children of participating parents is provided in Table 6.

Table 6

Frequency of Fundamental Cardiac Defect

Type	Frequency (%)
Atrial Septal Defect (ASD), Secundum	1 (3.2)
Ventricular Septal Defect (VSD), Type 2, Perimembraneous (Paramembranous) (Conoventricular)	4 (12.9)
Truncus arteriosus	2 (6.5)
Total anomalous pulmonary venous connection (TAPVC), Type 1 (supracardiac)	2 (6.5)
Total anomalous pulmonary venous connection (TAPVC), Type 2 (cardiac)	1 (3.2)
Tetralogy of Fallot (TOF)	1 (3.2)
Pulmonary atresia (PA), Intact ventricular septum (IVS)	1 (3.2)
Pulmonary atresia, VSD (Including TOF, PA)	2 (6.5)
Pulmonary stenosis, Valvar	1 (3.2)
Hypoplastic left heart syndrome (HLHS)	5 (16.1)
Single ventricle, Double Inlet Left Ventricle (DILV)	1 (3.2)
Single ventricle, Unbalanced AV canal	1 (3.2)
Transposition of Great Arteries (TGA), Intact Ventricular Septum (IVS)	2 (6.5)
Transposition of Great Arteries (TGA), Ventricular Septal Defect (VSD)	2 (6.5)
Double Outlet Right Ventricle (DORV), TGA type	1 (3.2)
Pulmonary artery sling	1 (3.2)
Interrupted aortic arch + VSD	2 (6.5)
Tetralogy of Fallot, Pulmonary stenosis	1 (3.2)

Note. ASD = Atrial septal defect; VSP = Ventricular septal defect; TAPVC = Total anomalous pulmonary venous connection; TOF = Tetralogy of Fallot; PA = Pulmonary atresia; IVS = Intact ventricular septum; HLHS = Hypoplastic left heart syndrome; DILV = Double inlet left ventricle; TGA = Transposition of the great arteries; DORV = Double outlet right ventricle.

Parent Demographics

Thirty-one parent pairs participated in this study. The mean age for mothers and fathers were 36.68, ±5.353 and 38.48, ±5.941, respectively. Race and ethnicity of the parent population was largely homogeneous, with mothers (n = 31, 90.3%) and fathers (n = 31, 93.5%) being of White race, and, of parents who reported ethnicity, mothers, (n = 19, 100%

White) and fathers, (*n* = 21, 54.8% White; 12.9% Hispanic or Latino). The mean education level for mothers and fathers was just under that of a bachelor's degree (mothers μ = 9.90, *SD*= 1.720) (fathers μ = 9.61, *SD* = 1.706). Parent pairs had a mean relationship length of 11.37 years.

Table 7

Parent Characteristics

Characteristics	Fathers Mean ± SD or Frequency (%)	Mothers Mean ± SD or Frequency (%)
Sample (*n*)	31	31
Age (Years)	38.48 ±5.941	36.68 ±5.353
Race		
White	29 (93.5)	28 (90.3)
Black or African American	0	1 (3.2)
Asian	1 (3.2)	1 (3.2)
Some other race	1 (3.2)	1 (3.2)
Ethnicity		
Hispanic or Latino origin (of any race)	4 (12.9)	0
White alone, not Hispanic or Latino	17 (54.8)	19 (61.3)
No Answer	10 (32.3)	12 (38.7)
Education Level		
Regular High School Diploma, GED or alternative	2 (6.4)	1 (3.2)
Some college credit, but less than 1 year of college credit	1 (3.2)	2 (6.5)
1 or more years of college credit, no degree	4 (12.9)	4 (12.9)
Associate degree	4 (12.9)	1 (3.2)
Bachelor's degree	12 (38.7)	11 (35.5)
Master's degree	5 (16.1)	8 (25.8)
Professional degree beyond a Bachelor's degree	12 (6.5)	3 (9.7)
Doctorate degree	2 (6.5)	1 (3.2)
Parent Type (Biological: Adoptive; Step/Bonus)	27 (87.1): 3 (9.7): 1 (3.2)	28 (90.3): 3 (9.7): 0

Households in this study had mean annual income falling within the $75,000-$99,999 range, a mean household size of 4.29 people, and a mean number of children per

household of 2.26. Only one family (3.2%) reported grandparents living in the home. The average length of the parent relationship was 11.37 (±3.9) years. See Table 8 for family demographics.

Table 8

Family Characteristics

Characteristics	Mean ± SD or Frequency (%)
Annual Household Income	
$35,000 to $49,999	4 (12.9)
$50,000 to $74,999	5 (16.1)
$75,000 to $99,999	7 (22.6)
$100,000 to $149,999	7 (22.6)
$150,000 to $199,999	1 (3.2)
$200,000 or greater	6 (19.4)
No Answer/ Unknown	1 (3.2)
Household Size	
3	10 (32.3)
4	9 (29.0)
5	7 (22.6)
6	3 (9.7)
7	2 (6.5)
Number of Children in Household	
1	10 (32.3)
2	10 (32.3)
3	6 (19.4)
4	3 (9.7)
5	2 (6.5)
Grandparents in Household (No: Yes)	30 (96.8): 1 (3.2)
Length of Relationship (Years)	11.37 ± 3.926

Data Analyses

In this study, one mother and one father from different households failed to complete the PEDsQL-FIM; therefore, analyses using variables from the PEDs-QL-FIM have a total sample size of 60; 30 mothers of a child with CHD (MCCHD), 30 fathers of a child with CHD (FCCHD), or 29 parental matched pairs. Data were initially checked for violation of

assumptions including the presence of normal distribution using normality plots with tests, homogeneity of variance using the Levene's test, and significance of skewness and kurtosis. Histograms and scatter plots were generated to visually assess for normal distribution and outliers of parent responses for each subscale and summary score by total parent population and by parent gender.

When analyzed as a single group or as gender-based subgroups, parent responses did not meet assumptions for normal distribution. Analyses to determine distribution characteristics and measures of variability included histograms, scatter plots, skewness and kurtosis. Box and whiskers plots were used to determine the presence of outliers due to the effect outliers can have on correlation testing (Morgan, Leech, Gloeckner, & Barrett, 2011). No outliers were found among parents within the PEDsQL-FIM subscales for summary scales; however, several PIP scales did have outliers for both genders. Outliers for mother reports occurred in the communication difficulty, role functioning frequency, role functioning difficulty, frequency total, and difficulty total subscales or summary scales. Both genders had an outlier present in the emotional distress frequency subscale (see Figure 4).

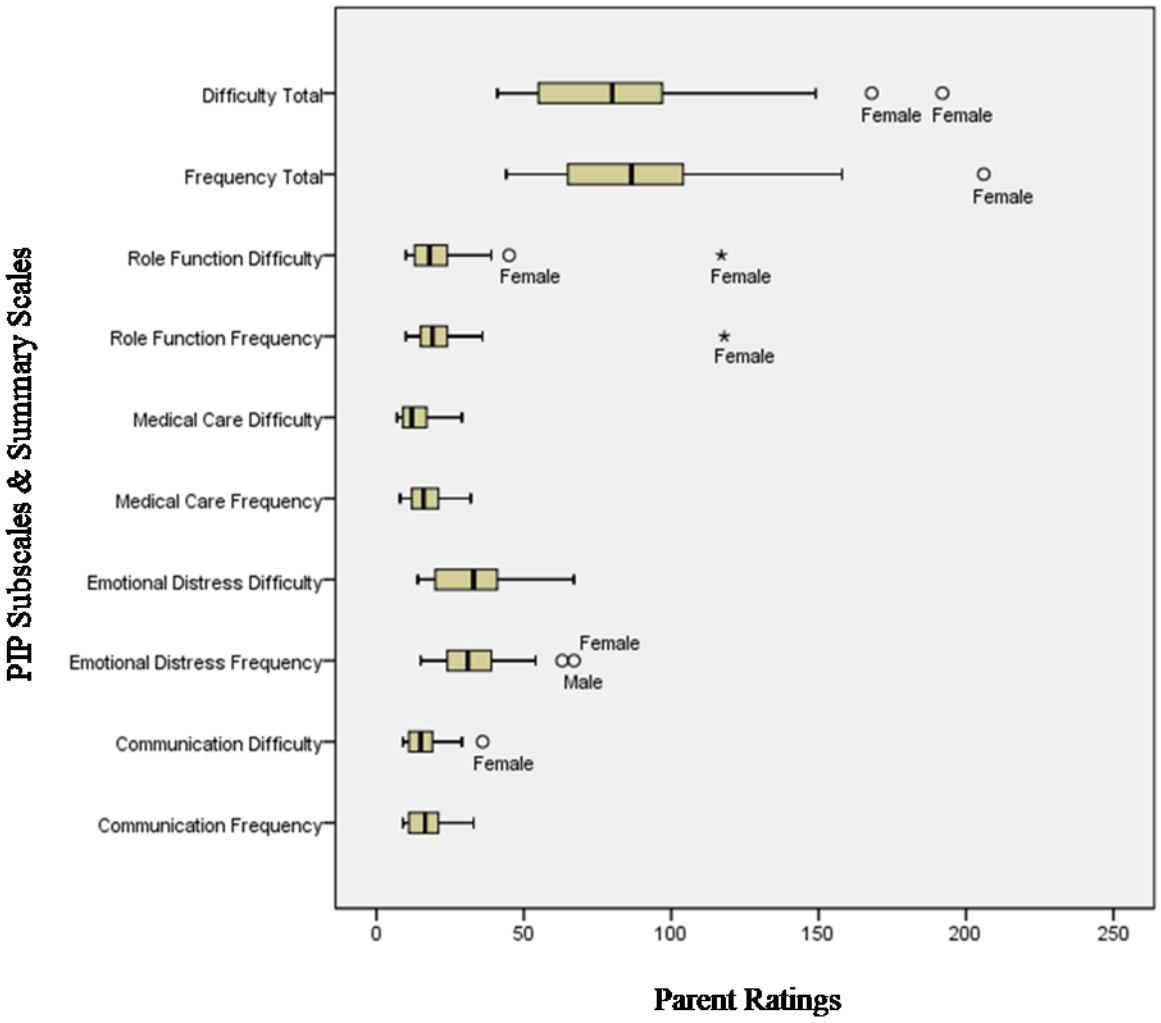


Figure 4. Box and whiskers plot demonstrating outliers in PIP scales.

Significant skewness to the right (≥ 1.0) was present in the following subscales or summary scales for all parents: communication difficulty (1.06), role function frequency (5.72), role difficulty (4.849); PIP frequency total (1.24), and PIP difficulty total (1.02). Fathers demonstrated slight skewness to the right (≥ 1.0) in the emotional distress frequency subscale (1.11) and emotional distress difficulty subscale (1.04) and slight skewness to the left (≤ -1.0) in PedsQL-FIM subscales of emotional functioning (-1.01), and social functioning (-1.61). Mothers demonstrated significant skewness to the right in the PIP role

function frequency (4.38), role function difficulty (3.82), and frequency total (1.24). Although Kurtosis values do not seem to affect results for most statistical analyses (Morgan et al., 2011), they were calculated during analysis of frequency distributions. Skewness and kurtosis scores for each subscale/summary scale and overall scores are displayed in Table 9.

Since assumptions were not met for normal distribution, there was a relatively small sample size of each parent subgroups (PIP n = 31 mothers/fathers, PedsQL-FIM n = 30 mothers/ fathers), and parent responses measure magnitude (therefore ordinal in nature), non-parametric tests were performed for all statistical analyses in this study.

For this study, main effects were calculated for each independent/ dependent relationship for all parents (PCCHD) as a group and for each gender-based subgroup when discussing specific scaled and summary scores. Gender-based comparisons were calculated to explore differences in the responses between fathers of a child with CHD (FCCHD) and mothers of a child with CHD (MCCHD) in each scale to determine if the difference between the parent genders was meaningful. Table 10 explains the score ranges for each subscale and summary scale to assist with understanding reported levels of stress, QOL, and family functioning.

Table 9

Skewness and Kurtosis

Subscale/ Summary Scale		Skewness			Kurtosis		
		PCCHD (n = 60)	FCCHD (n = 30)	MCCHD (n = 30)	PCCHD (n = 60)	FCCHD (n = 30)	MCCHD (n = 30)
PedsQL-FIM	Physical Functioning	-0.6	-0.5	-0.4	-0.7	-1.1	-1.0
	Emotional Functioning	-0.7	-1.0	-0.5	-0.6	0.3	-1.1
	Social Functioning	-1.0	-1.6	-0.6	-0.3	2.1	-1.1
	Cognitive Functioning	-0.4	-0.5	-0.2	-1.2	-1.2	-1.3
	Communication	-0.7	-1.2	-0.3	-0.7	1.1	-1.3
	Worry	-0.6	-0.8	-0.5	-0.5	0.1	-0.7
	Daily Activities	-0.7	-0.1	-0.6	-0.8	-1.2	-1.2
	Family Relationships	-0.8	-1.0	-0.7	-0.6	-0.01	-0.8
	QOL Summary	-0.5	-0.8	-0.3	-0.8	0.1	-1.3
	Family Functioning Summary	-0.6	-0.7	-0.7	-0.8	-0.7	-1.2
	Total	-0.7	-0.7	-0.3	-0.9	-0.1	-1.3
		PCCHD (n = 62)	FCCHD (n = 31)	MCCHD (n = 31)	PCCHD (n = 62)	FCCHD (n = 31)	MCCHD (n = 31)
PIP	Communications Frequency	0.4	0.3	0.4	-0.4	-1.0	-0.2
	Communications Difficulty	1.1	1.0	1.00	1.3	0.3	1.4
	Emotional Distress Frequency	0.9	1.1	0.7	1.0	2.0	0.7
	Emotional Distress Difficulty	0.5	1.0	0.1	-0.3	0.9	-0.5
	Medical Care Frequency	0.6	0.8	0.4	-0.7	0.1	-1.1
	Medical Care Frequency	0.6	0.8	0.4	-0.7	0.1	-1.1
	Medical Care Difficulty	0.9	0.6	0.9	0.3	-0.9	0.1
	Role Function Frequency	5.7	0.2	4.4	39.6	-0.7	22.0
	Role Function Difficulty	4.9	0.6	3.8	30.7	-0.2	17.9
	Frequency Total	1.2	-0.0	1.2	3.1	-0.9	2.2
	Difficulty Total	1.0	0.8	0.8	1.2	0.3	0.7

Note. PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Table 10

Meanings of PedsQL-FIM & PIP Scores

	Subscales and Summary Scales	Low QOL or family functioning	Moderate QOL or family functioning	High QOL or family functioning
PedsQL-FIM	Physical functioning	0-299	300-449	450-600
	Emotional functioning	0-249	250-374	375-500
	Social functioning	0-199	200-299	300-400
	Cognitive functioning	0-249	250-374	375-500
	Communication	0-149	150-225	225-300
	Worry	0-249	250-374	375-500
	Daily activities	0-149	150-225	225-300
	Family relations	0-249	250-374	375-500
	QOL summary	0-49	50-74	75-100
	Family functioning summary	0-49	50-74	75-100
	Total	0-49	50-74	75-100
		Low stress	Moderate stress	High stress
PIP	Communication frequency	9-18	19-35	36-45
	Communication difficulty	9-18	19-35	36-45
	Emotional distress frequency	15-30	31-59	60-75
	Emotional distress difficulty	15-30	31-59	60-75
	Medical care frequency	8-16	17-31	32-40
	Medical care difficulty	8-16	17-31	32-40
	Role functioning frequency	10-20	21-39	40-50
	Role functioning difficulty	10-20	21-39	40-50
	Frequency Total	42-84	85-167	168-210
	Difficulty Total	42-84	85-167	168-210

Results

Hypothesis 1 Findings

To investigate if there were statistically significant differences between mother and father perceptions of stress, QOL, and family functioning, Wilcoxon signed ranks with

Bonferroni correction (*comparison-wise alpha* = 0.025) were performed to compare the gender-based responses for each subscale/summary scale and overall scores. Wilcoxon signed ranks also demonstrated if the parent pairs from each household were aligned in their perceptions. One mother and one father, each from different families, failed to complete the PedsQL-FIM; therefore, only 29 parent pairs were included in the Wilcoxon matched pair analyses of PedsQL-FIM results. All parents completed the PIP; therefore 31 parent pairs were included in the matched pair analyses of the PIP results. The null hypothesis ($\alpha <$ 0.025) was rejected for Hypotheses 1a and 1b, because there were several statistically significant differences between mother and father reports in multiple measured areas of stress and QOL. The null hypothesis was accepted for H1c, since no statistically significant differences were noted among parent pair reports of family functioning.

Research Question 1a. Among parents of a child with CHD, what is the level of stress between mothers and fathers?

Hypothesis 1a. Mothers of a child with CHD will report higher levels of stress compared to fathers.

The hypothesis was accepted. Regarding parent reports of stress as measured by the PIP (N = 31 pairs), only role functioning difficulty was statistically significant (Z = -2.30, p = 0.02) where fathers (n = 31, *Mdn* = 16.00, *IQR* = 10) reported less difficulty in role functioning than mothers (n = 31, *Mdn* = 21.00, *IQR* = 16). All subscale and summary scale median scores for fathers fell within the low stress range except for stress frequency total, which fell within the moderate stress range. Mothers reported median scores in the low stress range for all subscales except for emotional distress frequency and difficulty, and

stress total frequency and total difficulty, where median scores fell within the moderate range (see Table 10).

Research Question 1b. Among parents of a child with CHD, what is the difference in the reporting of QOL perceptions between mothers and fathers?

Hypothesis 1b. Mothers of a child with CHD will report poorer QOL compared to fathers.

The hypothesis was accepted. Regarding QOL as measured by the PEDsQL-FIM subscales of physical, emotional, social, and cognitive functioning scores and HRQOL summary score, fathers generally reported higher scores for QOL than mothers; however, statistically significant scores were found in two areas ($N = 29$, $p < 0.025$). Fathers reported statistically significant ($Z = -2.52$, $p = 0.01$) better emotional functioning ($n = 29$, $Mdn = 450.00$, IQR = 162) in comparison to mothers ($n = 29$, $Mdn = 350.00$, IQR = 250). Fathers reported statistically significant ($Z = -2.38$, $p = 0.02$) better communication ($n = 29$, $Mdn = 275.00$, IQR = 100) in comparison to mothers ($n = 29$, $Mdn = 225.00$, IQR = 137.5). Fathers reported high levels of QOL in all subscales and summary scales, and mothers reported high levels of QOL in all subscales except emotional functioning, worry, and HRQOL summary scale, which were all in the range of moderate level of QOL.

Research Question 1c. Among parents of a child with CHD, what is the difference in the reporting of family functioning levels between mothers and fathers?

Hypothesis 1c. Mothers of a child with CHD will report lower levels of family functioning compared to fathers.

The null hypothesis was accepted. Regarding family functioning as measured by the PEDsQL-FIM family impact subscales of daily activities and family relationships, and

family functioning summary scale, there were no statistically significant contrasts between family functioning scores between mothers and fathers. Both mothers and fathers reported high levels of family functioning in all subscales (daily activities and family relations) and the family functioning summary scale.

Hypothesis 1 Summary

The gender-based effect size range (r = 0.01-0.50) of all measured scales and summary scores indicated small effect sizes; therefore, the differences between fathers and mothers of a child with CHD are not clinically meaningful. A larger sample size would assist in determining if these scores would reach levels of statistical significance. Using the effect sizes from this study, we determined 8,795 parent-pairs would be required for a fully powered (Power = 0.8) study. Wilcoxon matched pairs Z scores, significance, effect sizes, medians, interquartile ranges (IQR), levels of stress, QOL, and family functioning can be found on Table 11.

Table 11

Wilcoxon Matched Pairs Statistics

	Subscale / Summary Scale	Z score (sig. 2-tailed) (N = 29 pairs)	Effect size (N = 29 pairs)	Median (Interquartile Range (IQR)) FCCHD (n = 29)	Median (Interquartile Range (IQR)) MCCHD (n = 29)
PedsQL-FIM	Physical functioning	-1.7 (0.1)	0.4	500.0 (187.5) H	450.0 (250) H
	Emotional functioning	-2.5 (0.0)*	0.1	450.0 (162.5) H	350.0 (275) M
	Social functioning	-1.8 (0.1)	0.4	375.0 (100) H	325.0 (200) H
	Cognitive functioning	-0.4 (0.7)	0.1	375.0 (200) H	375.0 (200) H
	Communication	-2.4 (0.0)*	0.5	275.0 (100) H	225.0 (137.5) H
	Worry	-1.8 (0.1)	0.3	375.0 (200) H	350.0 (212.5) M
	Daily activities	-1.1 (0.3)	0.2	250.0 (125) H	250.0 (162.5) H
	Family relations	0.0 (1.0)	0.0	425.0 (200) H	475.0 (225) H
	QOL summary	-2.0 (0.1)	0.4	78.8 (29.4) H	71.3 (42.5) M
	Family functioning summary	-0.6 (0.5)	0.1	84.4 (42.1) H	84.4 (48.5) H
	Total	-1.9 (0.1)	0.4	80.6 (31.9) H	70.8 (40.6) M
		Z score (sig.2-tailed) (N = 31 pairs)	Effect size (N = 31 pairs)	FCCHD (n = 31)	MCCHD (n = 31)
PIP	Communication frequency	-1.0 (0.3)	0.3	16.0 (10) L	18.0 (9) L
	Communication difficulty	-2.0 (0.01)	0.5	13.0 (8) L	16.0 (9) L
	Emotional distress frequency	-1.6 (0.1)	0.3	30.0 (12) L	33.0 (18) M
	Emotional distress difficulty	-2.1 (0.0)	0.4	27.0 (17) L	37.0 (20) M
	Medical care frequency	-0.3 (0.8)	0.1	16.0 (8) L	16.0 (12) L
	Medical care difficulty	-1.1 (0.3)	0.3	12.0 (8) L	12.0 (10) L
	Role functioning frequency	-1.9 (0.1)	0.4	18.0 (7) L	20.0 (12) L
	Role functioning difficulty	-2.3 (0.0)*	0.4	16.0 (10) L	21.0 (16) L
	Frequency Total	-1.5 (0.1)	0.4	86.0 (35) M	86.0 (47) M
	Difficulty Total	-2.2 (0.0)	0.5	74.0 (36) L	86.0 (47) M

Note. *p < 0.025 level (2-tailed); **p < 0.005 level (2-tailed). L=Low, M=Moderate, High=High; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Hypothesis 2 Findings

To determine the influence specific aspects of stress and QOL have on family functioning, descriptive statistics and Spearman rho correlations were calculated to

determine if significant relationships existed between family functioning scaled score (dependent variable) and the remaining scales and summary scores (independent variables). Non-parametric tests (Spearman rho correlations) were completed for analysis. The null hypothesis was rejected ($\alpha < 0.05$), since all associations among scores of PedsQL-FIM QOL and PIP subscales and summary scales when compared with PedsQL-FIM family functioning summary scale were statistically significant ($p < 0.05$). Generally, family functioning was better in fathers ($\mu = 78.54$, $SD = 21.69$), who had a high level of functioning than mothers ($\mu = 74.48$, $SD = 26.12$), who had a moderate level of function.

Research Question 2a. Among parents of a child with CHD, what associations are present between parental perceptions of their stress and their family's functioning?

Hypothesis 2a. Parents who report high levels of stress will also report low family functioning levels.

The relationship of all parents' perspectives of their stress on their family's functioning is determined by comparing PIP subscale or summary scores with the PEDsQL-FIM family functioning summary score. Regarding the impact of stress on family functioning, correlations of PIP subscales and summary scores with the PedsQL-FIM family functioning summary score determined there were statistically significant relationships among all measured subscales and summary scales ($p < 0.05$), therefore, the null hypothesis was rejected. Fathers of a child with CHD reported lower mean scores in every PIP subscale and summary scale compared to mothers, indicating fathers perceived lower stress levels than mothers.

For all parents of a child with CHD, when compared with the PedsQL-FIM family functioning summary score, there were statistically significant negative correlations with

medium effect sizes (p = 0.00) in the PIP subscales of communication frequency, (r(58) = -0.65), and difficulty, (r(58) = -0.69); emotional distress frequency, (r(58) = -0.78), and difficulty, (r(58) = -0.64); medical care frequency (r(58) = -0.45), and difficulty, (r(58) = -0.53); role function frequency (r(58) = -0.70), and difficulty, (r(58) = -0.68); overall PIP frequency total (r(58) = -0.749), and difficulty total, (r(58) = -0.70). All resulting correlation coefficients were negative due to the inverse relationship in score reporting; high scores in the PIP indicate high stress levels and high scores in the PedsQL-FIM indicate high levels of QOL and family functioning.

In comparison to the family functioning summary score, nearly all PIP subscales and summary scales had medium effect sizes for all parents, ranging from (r(58) = -0.45 – 0.78). This suggests that parents who reported lower stress levels in any of the subscale or summary scale were highly likely to report better family functioning and vice versa. Among all parents, the lowest correlation coefficients were found when comparing stress experienced as part of the medical care frequency (r(58) = -0.45), and difficulty (r(58) = -0.53) of the child with CHD. This is suggestive that the perceived stress related to the health condition of the child with CHD is not as influential on the overall family's functioning as the other scales.

Research Question 2b. Among parents of a child with CHD, what associations are present between parental perceptions of their QOL with their family's functioning?

Hypothesis 2b. Parents who report poor QOL will also report low family functioning levels.

The null hypothesis was rejected. Regarding the impact of QOL on family functioning, correlations of PedsQL-FIM QOL subscales (physical, emotional, social and

cognitive functioning subscales) and QOL summary scores with the PedsQL-FIM family functioning summary score indicated statistically significant relationships among all measured subscales and summary scales ($p = 0.00$). The strongest gender-based associations with family functioning were found between emotional functioning where fathers ($\mu = 400.83$, $SD = 111.51$) reported higher levels of QOL than mothers ($\mu = 338.33$, $SD = 132.89$), ($r(58) = 0.76$, $p=0.00$), and social functioning where fathers ($\mu = 330.00$, $SD = 95.46$) reported high levels of QOL compared to the moderate levels of QOL among mothers ($\mu = 284.17$, $SD = 118.45$), *(*$r(58) = 0.83$, $p = 0.00$).

Large effect sizes were noted among each variable correlation (all $p = 0.00$). For all parents, when compared with the PedsQL-FIM family functioning summary score, physical functioning ($r(58) = 0.73$), emotional functioning ($r(58) = 0.76$), social functioning subscale ($r(58) = 0.83$), cognitive functioning subscale *(*$r(58) = 0.69$), were found to have large effect sizes when correlated with the family functioning summary score. The PedsQL-FIM QOL summary score and family functioning summary scores are positively correlated with a large effect size, ($r(58) = 0.84$, $p = 0.00$).

Hypothesis 2 Summary

The descriptive statistics for scaled and summary scores indicated that parents who report better outcomes in their stress and QOL also report better overall family functioning and vice versa. Parents who experienced better sleep, fewer headaches, periods of their own illness, felt energetic, and were better able to take physical care of themselves reported better family functioning. Parents who did not feel isolated and had time to foster social relationships reported better family functioning. Regarding communication-related stressors, parents who reported less frequency and difficulty in communication stressors such as

arguing with family members or speaking with care providers or with their child about their illness tended to report better overall family functioning and vice versa. Parents who reported less frequent or difficult occurrences of emotional distress such as feeling numb inside, depressed, or hopeless, or learning upsetting news tended to report better overall family functioning and vice versa. Parents who reported less stress involving the medical care matters of their child with CHD such as assisting with their child's medical procedures, navigating changes to their child's medical routines or needs, fewer hospital stays, or having to make decisions about their child's medical care reported better family functioning. Parents who reported more stress in the frequency and difficulty of their role functioning, (such as struggling with the conflict of going to work to provide financially instead of remaining present with their child who is ill, or struggling with the discipline of their child with CHD who may have behavior challenges associated with adverse neurological sequalae resulted from their CHD type or course of care) reported lower family functioning. Descriptive statistics for all measures can be found in Table 12.

Table 12

Descriptive Statistics of Parent Measures

Subscale / Summary Scale		Mean			Standard Deviation		
		PCCHD (n = 60)	FCCHD (n = 30)	MCCHD (n = 30)	PCCHD (n = 60)	FCCHD (n = 30)	MCCHD (n = 30)
PedsQL-FIM	Physical Functioning	452.5 H	476.7 H	428.3 M	130.2	112.9	143.2
	Emotional Functioning	369.6 M	400.8 H	338.3 M	125.6	111.5	132.9
	Social Functioning	307.1 H	330.0 H	284.2 M	109.1	95.5	118.5
	Cognitive Functioning	385.0 H	386.7 H	383.3 H	109.8	121.0	99.4
	Communication	225.4 H	244.2 H	206.7 M	73.7	66.5	76.8
		355.4 M	377.5 H	333.3 M	120.34	117.9	120.8
	Daily Activities	222.5 M	235.0 H	210.0 M	79.4	67.5	89.2
	Family Relationships	389.6 H	393.3 H	385.8 H	123.1	117.6	130.3
	QOL Summary	75.7 H	79.7 H	71.7 M	21.5	19.0	23.3
	Family Functioning Summary	76.5 H	78.5 H	74.5 M	23.9	21.7	26.1
	Total	75.2 H	79.00H	71.4 M	20.8	18.6	22.5
		All (n = 62)	Fathers (n = 31)	Mothers (n = 31)	All (n = 62)	Fathers (n = 31)	Mothers (n = 31)
PIP	Communications Frequency	17.15 L	16.1 L	18.2 L	6.0	5.6	6.2
	Communications Difficulty	15.74 L	14.4 L	17.3 L	5.9	5.1	6.4
	Emotional Distress Frequency	32.4 M	30.6 L	34.3 M	11.1	10.3	11.6
	Emotional Distress Difficulty	32.6 M	29.8 L	35.4 M	13.2	12.5	1367
	Medical Care Frequency	17.0 M	16.6 L	17.5 M	6.5	6.0	7.1
	Medical Care Difficulty	13.5 L	12.6 L	14.3 L	5.3	4.6	6.0
	Role Function Frequency	21.2 M	18.1 L	24.4 M	13.9	4.5	18.8
	Role Function Difficulty	21.1 M	17.4 M	24.8 M	14.6	5.6	19.4
	Frequency Total	87.8 M	81.4 L	94.3 M	29.2	21.8	34.8
	Difficulty Total	82.9 L	74.2 L	91.6 M	32.7	25.4	37.1

Note. PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease. L=Low stress, QOL, or family functioning; M= Moderate stress, QOL, or family functioning; H= High stress, QOL, or family functioning.

Spearman rho statistics for all parents and gender-based subgroups when compared to family functioning can be found in Table 13.

Table 13

Associations of QOL and Stress with Family Functioning

	Subscale / Summary Scale	Spearman rho statistic (Sig.)		
		PCCHD (N = 60)	FCCHD (n = 30)	MCCHD (n = 30)
PEDsQL-FIM	Physical functioning	0.7** (0.0)	0.7** (0.0)	0.7** (0.0)
	Emotional Functioning	0.8** (0.0)	0.7** (0.0)	0.8** (0.0)
	Social functioning	0.8** (0.0)	0.8** (0.0)	0.9** (0.0)
	Cognitive functioning	0.7** (0.0)	0.6** (0.0)	0.8** (0.0)
	Communication	0.7** (0.0)	0.7** (0.0)	0.7** (0.0)
	Worry	0.7** (0.0)	0.8** (0.0)	0.7** (0.0)
	Daily activities	0.9** (0.0)	0.9** (0.0)	0.9** (0.0)
	Family relations	1.0** (0.0)	1.0** (0.0)	0.9** (0.0)
	QOL summary	0.8** (0.0)	0.8** (0.0)	0.9** (0.0)
	Total	0.9** (0.0)	0.9** (0.0)	0.9** (0.0)
PIP	Communication frequency	-0.7** (0.0)	-0.67** (0.0)	-0.6** (0.0)
	Communication difficulty	-0.7** (0.0)	-0.8** (0.0)	-0.6** (0.0)
	Emotional distress frequency	-0.8** (0.0)	-0.8** (0.0)	-0.7** (0.0)
	Emotional distress difficulty	-0.6** (0.0)	-0.7** (0.0)	-0.5** (0.0)
	Medical care frequency	-0.5** (0.0)	-0.4* (0.0)	-0.5** (0.0)
	Medical care difficulty	-0.5** (0.0)	-0.6** (0.0)	-0.4* (0.0)
	Role functioning frequency	-0.7** (0.0)	-0.8** (0.0)	-0.7** (0.0)
	Role functioning difficulty	-0.7** (0.0)	-0.7** (0.0)	-0.6** (0.0)
	Frequency Total	-0.7** (0.0)	-0.8** (0.0)	-0.7** (0.0)
	Difficulty Total	-0.7** (0.0)	-0.8** (0.0)	-0.6** (0.0)

Note. *$p < 0.05$ level (2-tailed); **$p < 0.01$ level (2-tailed). PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Hypothesis 3 Findings

To investigate the impact the severity of the child's CHD diagnosis has on parental perceptions of stress, QOL, and family functioning, a Spearman rho correlation matrix was created to explore for statistically significant relationships between the PEDsQL-FIM and

PIP scaled and summary scores and the CHD severity (STAT) score derived from the STS-EACTS congenital heart surgery mortality (STAT) scoring system (Jacobs et al., 2012; O'Brien et al., 2009). Scores ranging from 0.1 to 5.0 were assigned to a surgical procedure based its estimated mortality and, based upon increasing risk, categorized into five homogeneous categories (O'Brien et al., 2009). For the sake of this study, the highest STAT score a child had received between birth and date of neurodevelopmental testing was used to measure complexity/severity of fundamental cardiac defect (CHD).

There were no statistically significant associations between parental stress, QOL, and family functioning scales and summary scores and severity of CHD type as measured by the STAT score; therefore, the null hypothesis was accepted ($\alpha = 0.05$).

Hypothesis 3 Summary

The results indicated that parent perceptions of their stress, QOL or family's functioning were not significantly impacted by the severity of the child's heart disease. Mean, standard deviation, Spearman correlation coefficient, and significance for all parents, fathers and mothers of a child with CHD can be found in Table 14.

Table 14

Parental Outcomes in Relation to CHD Severity

	Subscale / Summary Scale	Spearman rho statistic (significance)		
		PCCHD (n = 60)	FCCHD (n = 30)	MCCHD (n = 30)
PedsQL-FIM	Physical Functioning	-0.0 (1.0)	-0.0 (1.0)	0.0 (0.8)
	Emotional Functioning	0.0 (0.7)	0.1 (0.8)	0.1 (0.6)
	Social Functioning	0.1 (0.6)	0.1 (0.5)	0.1 (0.8)
	Cognitive Functioning	0.2 (0.2)	0.2 (0.4)	0.1 (0.5)
	Communication	-0.0 (0.9)	0.1 (0.6)	-0.1 (0.8)
	Worry	-0.2 (0.1)	-0.3 (0.2)	-0.1 (0.5)
	Daily Activities	-0.0 (0.8)	-0.1 (0.7)	0.0 (1.0)
	Family Relations	-0.0 (0.7)	-0.0 (0.9)	-0.1 (0.7)
	QOL Summary	0.0 (0.8)	0.1 (0.7)	0.1 (0.8)
	Family Functioning Summary	-0.0 (0.8)	-0.1 (0.8)	-0.0 (0.9)
	Total	-0.0 (0.8)	-0.2 (0.9)	-0.0 (1.0)
		PCCHD (n = 62)	FCCHD (n = 31)	MCCHD (n = 31)
PIP	Communication Frequency	0.1 (0.4)	0.1 (0.6)	0.1 (0.6)
	Communication Difficulty	-0.0 (0.9)	-0.1 (0.7)	0.0 (0.7)
	Emotional Distress Frequency	0.0 (0.8)	0.1 (0.5)	-0.1 (0.6)
	Emotional Distress Difficulty	-0.1 (1.0)	0.1 (0.6)	-0.2 (0.3)
	Medical Care Frequency	0.1 (0.4)	0.1 (0.7)	0.1 (0.6)
	Medical Care Difficulty	-0.2 (0.2)	-0.2 (0.2)	-0.2 (0.4)
	Role Functioning Frequency	0.0 (1.0)	0.1 (0.7)	-0.0 (0.8)
	Role Functioning Difficulty	-0.2 (0.3)	-0.2 (0.4)	-0.2 (0.4)
	Frequency Total	0.1 (0.7)	0.1 (0.5)	-0.0 (0.9)
	Difficulty Total	-0.7 (0.6)	-0.0 (0.9)	-0.1 (0.5)

Note. *$p < 0.05$ level (2-tailed)'; **$p < 0.01$ level (2-tailed) PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Additional Analyses

Influence of Time on Parent Perspectives

A Spearman rho correlation matrix was created to explore for statistically significant relationships between the PIP and PEDsQL-FIM scaled and summary scores and times since first and most recent surgery requiring cardio-pulmonary bypass. Time since most recent

CBP surgery had a positive association on communication of all parents ($r(58) = 0.275$, $p = 0.03$), and fathers of a child with CHD *(*$r(28) = 0.396$, $p = 0.03$). There were no significant relationships between time elapsed and measured scales for mothers. This indicates that for all parents and fathers, their stress related to communication functioning with others regarding their child's condition improved as more time passed since their child's most recent CPB surgery. Time since child's first CPB surgery had a negative, and statistically significant association with medical care difficulty for all parents ($r(58) = -0.286$, $p = 0.02$). This indicates that the more elapsed time since the child's first surgery, the less difficulty parents reported in providing medical care such as assisting with medical procedures, making decisions about their child's medical care, or managing changes in their child's medical care routines. Correlations for all measured susbscales/summary scales with elapsed time since their child's first and most recent CPB surgery can be seen in Table 15.

Table 15

Parental Outcomes in Relation to Elapsed Time since Child's CPB Surgeries

	Subscale / Summary Scale	Spearman rho statistic (significance)					
		PCCHD (n = 60)		FCCHD (n = 30)		MCCHD (n = 30)	
		First	Most recent	First	Most recent	First	Most recent
PedsQL-FIM	Physical Functioning	0.0 (0.9)	0.2 (0.2)	0.0 (0.9)	0.3 (0.1)	0.0 (1.0)	0.0 (0.8)
	Emotional Functioning	-0.0 (0.8)	0.1 (0.3)	-0.1 (0.6)	0.3 (0.2)	0.0 (0.9)	0.0 (1.0)
	Social Functioning	0.1 (0.4)	0.2 (0.1)	0.1 (0.6)	0.3 (0.1)	0.1 (0.6)	0.1 (0.5)
	Cognitive Functioning	-0.0 (0.9)	0.0 (0.9)	-0.1 (0.8)	0.0 (1.0)	0.0 (1.0)	0.0 (0.9)
	Communication	0.1 (0.6)	0.3* (0.0)	0.1 (0.5)	0.4* (0.0)	0.0 (1.0)	0.1 (0.5)
	Worry	-0.1 (0.3)	0.12 (0.2)	-0.1 (0.5)	0.2 (0.2)	-0.2 (0.4)	0.1 (0.6)
	Daily Activities	0.0 (0.9)	0.2 (0.2)	-0.1 (0.8)	0.2 (0.3)	0.1 (0.8)	0.1 (0.5)
	Family Relations	-0.1 (0.7)	0.01 (0.7)	-0.1 (0.6)	0.09 (0.6)	-0.0 (1.0)	0.0 (0.9)
	QOL Summary	0.0 (1.0)	0.2 (0.3)	-0.0 (0.9)	0.3 (0.2)	0.0 (0.9)	0.1 (0.7)
	Family Functioning Summary	-0.0 (0.8)	0.0 (0.6)	-0.1 (0.7)	0.1 (0.5)	-0.01 (1.0)	0.0 (0.3)
	Total	-0.0 (0.3)	0.1 (0.3)	-0.1 (0.7)	0.2 (0.2)	0.00 (1.0)	0.1 (0.7)
		PCCHD (n = 62)		FCCHD (n = 31)		MCCHD (n = 31)	
		First	Most recent	First	Most recent	First	Most recent
PIP	Communication Frequency	0.1 (0.7)	0.01 (0.9)	0.1 (0.7)	-0.0 (0.9)	0.0 (1.0)	0.03 (0.9)
	Communication Difficulty	0.1 (0.7)	0.04 (0.7)	0.0 (1.0)	0.0 (1.0)	0.1 (0.6)	0.06 (0.7)
	Emotional Distress Frequency	-0.1 (0.7)	-0.17 (0.2)	-0.0 (0.8)	-0.3 (0.2)	-0.1 (0.7)	-0.08 (0.7)
	Emotional Distress Difficulty	0.0 (1.0)	-0.08 (0.5)	0.0 (0.9)	-0.1 (0.5)	-0.1 (0.7)	-0.1 (0.7)
	Medical Care Frequency	-0.2 (0.1)	-0.17 (0.2)	-0.2 (0.3)	-0.2 (0.4)	-0.3 (0.2)	-0.2 (0.3)
	Medical Care Difficulty	-0.3* (0.0)	-0.05 (0.7)	-0.3 (0.1)	-0.0 (0.6)	-0.3 (0.3)	-0.0 (0.8)
	Role Functioning Frequency	-0.1 (0.6)	-0.05 (0.7)	-0.2 (0.4)	-0.2 (0.3)	-0.0 (1.0)	0.1 (0.8)
	Role Functioning Difficulty	-0.2 (0.3)	-0.1 (0.6)	-0.2 (0.4)	-0.2 (0.3)	-0.2 (0.4)	0.0 (1.0)
	Frequency Total	-0.1 (0.6)	-0.1 (0.4)	-0.0 (0.9)	-0.2 (0.3)	-0.1 (0.6)	-0.1 (0.8)
	Difficulty Total	-0.1 (0.5)	-0.1 (0.6)	-0.1 (0.6)	-0.1 (0.5)	-0.1 (0.5)	-0.1 (0.8)

Note. *p < 0.05 level (2-tailed); **p < 0.01 level (2-tailed). PCCHD = Parents of child with congenital heart disease; FCCHD = Fathers of child with congenital heart disease; MCCHD = Mothers of child with congenital heart disease.

Effect of Cardiac Surgery Frequency

The secondary sample had children who had up to four CBP surgeries. To examine for significant effect of the number of CBP surgeries their child has experienced on parent reports, Kruskal-Wallis nonparametric tests were performed. The Kruskall-Wallis nonparametric test showed no statistically significant differences among scores of mothers dependent upon the number of CBP surgeries their child had experienced. However, the test indicated significant differences for fathers in social functioning (χ^2 (3, N = 30, 9.39, p = 0.03)) , cognitive functioning (χ^2 (3, N = 30, 8.61, p = 0.04)), PedsQL-FIM HRQL summary (χ^2 (3, N = 30, 8.35, p = 0.04)), role function frequency (χ^2 (3, N = 31, 9.60, p = 0.02)), and role functioning difficulty (χ^2 (3, N = 31, 9.10, p = 0.03)). Post hoc Mann-Whitney tests compared the number of CBP surgeries on these scores using a Bonferroni corrected p value of 0.008 to indicate statistical significance. The only statistically significant differences were found in role function frequency and role function difficulty for fathers. Role function frequency was significantly higher in fathers whose child had four (n = 4, Mdn = 25.00, IQR = 4) CBP surgeries than those who child had only one (n = 14, m Mdn = 17.00, IQR = 7) CBP surgery (U = 3.00, p = 0.007, r = -0.63), a medium effect size (Leech, Barrett, & Morgan, 2011). Role function difficulty was significantly higher in fathers whose child had four (n = 4, Mdn = 24.00, IQR = 8) CBP surgeries than those who child had only two (n = 7, Mdn = 15.00, IQR = 4) CBP surgeries (U =0.00, p = 0.008, r = -0.80), a large effect size (Leech et al., 2011). This demonstrates fathers whose child had had more CBP surgeries report worse perspectives about their ability to fulfill their role as the family's financial provider, father, or spouse.

Abnormal Brain Imaging Effect

For all parents of a child with CHD , numerous statistically significant associations among PedsQL-FIM scores with small effects were noted when a child had abnormal brain imaging: physical function ($r(58) = -0.296$, $p = 0.02$); social functioning ($r(58) = -0.254$, $p = 0.05$); worry ($r(58) = -0.281$, $p = 0.03$); daily activities ($r(58) = -0.314$, $p = 0.01$); parent HRQL summary score ($r(58) = -0.260$, $p = 0.04$); family functioning summary score, ($r(58) = -0.260$, $p = 0.05$); and total FIM score ($r(58) = -0.267$, $p = 0.04$). This indicates that having a child with CHD and abnormal brain imaging, negatively influences many aspects of their parents' QOL and family functioning. No statistically significant associations were found among reported PIP scores. Correlations for PedsQL-FIM subscales/summary scales with abnormal brain imaging can be seen in Table 16.

Table 16

Correlations between Parent Measures with Abnormal Brain Imaging

	Subscale / Summary Scale	PCCHD Correlation Coefficient (Sig.) $n = 60$	FFCHD Correlation Coefficient (Sig.) $n = 30$	MCCHD Correlation Coefficient (Sig.) $n = 30$
PedsQL-FIM	Physical Functioning	-0.3* (0.0)	-0.3 (0.1)	-0.2 (0.2)
	Emotional Functioning	-0.1 (0.4)	-0.2 (0.3)	-0.1 (0.8)
	Social Functioning	-0.3* (0.1)	-0.3 (0.2)	-0.3 (0.1)
	Cognitive Functioning	-0.2 (0.1)	-0.1 (0.4)	0.2 (0.2)
	Communication	-0.2 (0.2)	-0.2 (0.3)	-0.1 (0.5)
	Worry	-0.3* (0.0)	-0.4* (0.0)	-0.2 (0.3)
	Daily Activities	-0.3* (0.0)	-0.2 (0.2)	-0.2 (0.2)
	Family Relationships	-0.3 (0.1)	-0.2 (0.4)	-0.3 (0.1)
	Parent HRQL Summary Score	-0.3* (0.0)	-0.3 (0.1)	-0.3 (0.2)
	Family Functioning Summary Score	-.03* (0.0)	-0.2 (0.3)	-0.3 (0.1)
	Total FIM Score	-0.3* (0.0)	-0.3 (0.1)	-0.3 (0.2)
		PCCHD ($n = 62$)	FCCHD ($n = 31$)	MCCHD ($n = 31$)
PIP	Communication Frequency	0.1 (0.7)	0.0 (0.9)	0.1 (0.6)
	Communication Difficulty	0.0 (1.0)	0.0 (0.8)	0.0 (1.0)
	Emotional Distress Frequency	0.2 (0.3)	0.2 (0.2)	0.1 (0.6)
	Emotional Distress Difficulty	0.1 (0.4)	0.1 (0.5)	0.1 (0.6)
	Medical Care Frequency	0.28 (0.2)	0.1 (0.5)	0.2 (0.3)
	Medical Care Difficulty	0.1 (0.7)	0.0 (0.9)	0.1 (0.7)
	Role Functioning Frequency	0.2 (0.3)	0.1 (0.5)	0.1 (0.5)
	Role Functioning Difficulty	0.1 (0.5)	0.2 (0.4)	0.1 (0.8)
	Frequency Total	0.2 (0.3)	0.2 (0.3)	0.1 (0.5)
	Difficulty Total	0.1 (0.5	0.1 (0.4)	0.1 (0.7)

Note. *$p < 0.05$ level (2-tailed); **$p < 0.01$ level (2-tailed). PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Early Intervention Services Effect

Several statistically significant associations were found between parent responses and whether their child had received early intervention services during infant/toddler years. Early interventions are services and supports for babies and toddlers with identified developmental delays or differences (Center for Parent Information and Resources, 2017). Statistically significant associations were noted among parents whose child had received

early intervention services and their reports of less worry and/or better daily activities: for all parents, ($r(58) = -0.281$, $p = 0.03$); daily activities ($r(58) = -0.328$, $p = 0.01$); for fathers, worry ($r(29) = -0.374$, $p = 0.04$); and for mothers, daily activities ($r(29) = -0.393$, $p = 0.03$). Correlations for PedsQL-FIM and PIP subscales and summary scales with early intervention services can be seen in Table 17.

Table 17

Correlations between Parent Measures with Early Intervention Services

	Subscale / Summary Scale	PCCHD Correlation Coefficient (Sig.) $n = 60$	FCCHD Correlation Coefficient (Sig.) $n = 30$	MCCHD Correlation Coefficient (Sig.) $n = 30$
PedsQL-FIM	Physical Functioning	-0.2 (0.1)	-0.3 (0.1)	-0.2 (0.4)
	Emotional Functioning	-0.2 (0.2)	-0.3 (0.1)	-0.1 (0.8)
	Social Functioning	-0.2 (0.1)	-0.3 (0.1)	-0.1 (0.5)
	Cognitive Functioning	-0.1 (0.4)	-0.1 (0.5)	-0.1 (0.6)
	Communication	-0.1 (0.6)	-0.3 (0.2)	0.1 (0.6)
	Worry	-0.3* (0.0)	-0.4* (0.0)	-0.2 (0.3)
	Daily Activities	-0.3* (0.0)	-0.3 (0.2)	-0.4* (0.0)
	Family Relationships	-0.2 (0.2)	-0.2 (0.4)	-0.2 (0.3)
	Parent HRQL Summary Score	-0.2 (0.1)	-0.3 (0.1)	-0.1 (0.5)
	Family Functioning Summary Score	-0.2 (0.1)	-0.2 (0.3)	-0.3 (0.2)
	Total FIM Score	-0.2 (0.1)	-0.3 (0.1)	-0.17 (0.4)
		PCCHD ($n = 62$)	FCCHD ($n = 31$)	MCCHD ($n = 31$)
PIP	Communication Frequency	0.1 (0.7)	0.0 (0.9)	0.1 (0.6)
	Communication Difficulty	0.0 (1.0)	0.0 (0.8)	0.0 (1.0)
	Emotional Distress Frequency	0.2 (0.3)	0.2 (0.2)	0.1 (0.6)
	Emotional Distress Difficulty	0.1 (0.4)	0.1 (0.5)	0.1 (0.6)
	Medical Care Frequency	0.2 (0.2)	0.1 (0.5)	0.2 (0.3)
	Medical Care Difficulty	0.1 (0.7)	0.0 (0.9)	0.1 (0.7)
	Role Functioning Frequency	0.2 (0.3)	0.1 (0.5)	0.1 (0.5)
	Role Functioning Difficulty	0.1 (0.5)	0.2 (0.4)	0.1 (0.8)
	Frequency Total	0.2 (0.3)	0.2 (0.3)	0.1 (0.5)
	Difficulty Total	0.1 (0.5)	0.1 (0.4)	0.1 (0.7)

Note. *$p < 0.05$ level (2-tailed); **$p < 0.01$ level (2-tailed). PCCHD= Parents of child with congenital heart disease; FCCHD= Fathers of child with congenital heart disease; MCCHD= Mothers of child with congenital heart disease.

Response to Acute Parental Scores

The PI collaborated with the Heart Center's Thrive program to develop a response in the event parent participants endorsed acute levels of stress, poor QOL, or family functioning. The Thrive program is designed to provide support and resources for families and patients served by the Ward Family Heart Center. Participating parents that endorsed grand total scores for either measure in the appropriate quartile of test score ranges to indicate most negatively impacted stress, QOL, and family functioning (≥336 PIP, ≤ 900 PEDs QL-FIM) were provided information about the Heart Center's Thrive program. One family was sent Thrive program information due to the mother's total PIP responses (frequency total + difficulty total = 398) in the highest quartile (≥336), indicating very high stress.

This chapter discussed the participation response rates, sample demographics, preliminary analyses for assumption testing, and findings for each proposed hypothesis and additional analyses performed. The next chapter provides an overview of the research questions posed in this study and includes a discussion of the evolution of research on parents of a child with CHD, a synthesis and contextualization of the study's major findings with previous research related to parental outcomes when having a child with CHD or other chronic disease. Strengths and limitations of the PinCHeD study are reviewed as well as the implications related to the findings.

CHAPTER 5

DISCUSSION

An overview of the results of this study is provided, followed by a discussion describing how this study compares or contrasts with previous research on parental perspectives when having a child with CHD. A section is included to focus specifically on the differences in methodology and findings of the PinCHeD study related to stress, QOL, and family functioning in context with three recent studies that also utilized the PIP as an instrument to assess parental stress (Bishop et al., 2019; Caris et al., 2016; Kaugars, Shields, & Brosig, 2018). Strengths and limitations of the PinCHeD study are reviewed as well as the implications in areas of practice, theory, research, and policy. Recommendations specific to practice, theory, research, and policy related to parents of a child with CHD are described, followed by the conclusion.

Overview of Findings

The main findings of this study demonstrated that no statistically significant differences exist between mothers and fathers in their perceptions of stress, QOL, or family functioning when having a child with CHD. Parents reported reciprocal relationships in the outcomes regarding their stress levels, QOL, and family functioning. For example, parents who reported low stress levels also reported higher QOL and better family functioning. Severity of CHD diagnosis for the child was not associated with higher parental stress, lower QOL, or poor family functioning. Parents reported better perceptions of the stress, QOL, and family functioning as more time passed since their child's cardiac surgeries, demonstrating bonadaption and adjustment to their situation over time. Statistically significant correlations were present only among fathers in their reports of elevated intensity and difficulty in their

role functioning associated with the higher number of cardiac surgeries their child had undergone. Having a child with CHD and abnormal brain imaging negatively influenced many aspects of parents' QOL and family functioning. Parents whose child received early intervention services felt less anxiety and concern over their child's future or how other family members were affected by the medical condition of the child with CHD.

Contextualization of Findings with Previous Research

This study did not reflect the high rates of acute levels of stress or poor QOL and lower family functioning as previously reported in research participants of parents of a child with CHD (Gregory et al., 2018). This could be due to the selective sampling methods used to recruit in those studies. The lack of clinically significant levels of stress among the PinCHeD study's sample is consistent with the findings of other recent studies on parents of a child with CHD (Caris et al., 2016; Kaugars et al., 2018). For example, Caris et al. (2016) studied parents of a child with hypoplastic left heart syndrome and Kaugar et al.'s (2018) sample was comprised of parents with identified concerns for their child with CHD warranting a referral for follow up with a pediatric psychologist (nearly 60% of which had single ventricle pathophysiology). The addition of psychological or developmental differences in their sample of children with severe forms of CHD may be a compounding factor for adverse parental stress and QOL. These specific sample characteristics limit application to broader populations.

Parents in the PinCHeD study who had a child with CHD and who had received early intervention services (indicating developmental delays or differences identified in the child under three years of age) reported significant levels of anxiety and concern over their child's future or how other family members were affected by the medical condition of the

child with CHD. In regard to the effects of CHD severity on parental outcomes, the PinCHeD sample was comprised of 25% parents of a child with single ventricle pathophysiology or highest CHD severity score of five. Correlations between aggregate scales measuring parental stress, QOL, and family functioning in the PinCHeD study were similar to those reported in the Kaugars et al. (2018) study even though the child's CHD types were collectively not as severe. Caris (2016) looked specifically at parents of children with hypoplastic left heart syndrome, and, although Caris's mean values were higher in each PIP scaled score than this study, the findings were similar in that neither study demonstrated clinically significant levels of parental stress, negatively impacted QOL, or lower levels of family functioning. Both the Caris (2016) study and the PinCHeD study reported correlations suggesting that high QOL was associated with lower stress levels and higher levels of family functioning. The implications of these findings are first, CHD type or severity was not an accurate predictor of adverse parental outcomes. Second, although higher than parents of healthy children, stress, QOL, and family functioning were not at clinically significant levels among parents of a child with CHD. Lastly, due to the correlations among stress, QOL and family functioning, support that improves parental QOL may also improve the parent's ability to more effectively manage stressful experiences, which improves overall family functioning.

There are differences in the reporting of the effects of gender on parental outcomes when having a child with CHD in the recent CHD literature. Gender-based parent outcomes were not reported in the Kaugars et al. (2018) study, and the Caris (2016) study was comprised of 87% mothers. These two studies purposively selected for CHD type or known developmental or behavioral differences requiring psychology follow up; however, the

PinCHeD study purposively selected for parent dyads for equal, gender-based subgroups to determine if mothers and fathers responded differently when asked about their experiences and perceptions of their own QOL and their family's functioning.

In the PinCHeD study, all parents of a child with CHD reported higher scores related to worry about their child's future compared to other questions or measured scales addressing aspects of emotional distress such as helplessness, anxiety, or fear. This is consistent with previous literature. For example, numerous studies examining psychological experiences of parents of a child with CHD have demonstrated higher emotional distress, such as anxiety, stress, and feelings of being numb or isolated and were higher than parents of healthy children or children with other chronic health conditions (Bevilacqua et al., 2013; Diffin, Spence, Naranian, & Badawi, 2016; Ezzat et al., 2016; Utens et al., 2000). The PinCHeD study found mothers typically report higher emotional dysfunction and more worry than fathers suggesting that the time elapsed since their child's cardiac surgery may be a factor in the intensity and frequency that stress and emotional distress are experienced.

The parents in this study did not feel isolated or experience a lack support. This contrasts with findings of earlier research where parents of a child with CHD reported high levels of loneliness, isolation, and decreased support (Diffin et al., 2016; Doherty et al., 2009; Levert, Helbing, Dulfer, van Domburg, & Utens, 2016). Parents in the current study did not report difficulty in finding time or feeling up to participating in social activities. This demonstrates a level of bonadaption in which the participating parents may have learned to take opportunities to meet life values or needs such as leisure activities that foster social activities, positive emotions, and the development of new skills and interests (Brajša-Žganec, Merkaš, & Šverko, 2011). Although social expectations generally place mothers as

the main care provider for the children in the family, the father's role and self-identification as the family's breadwinner may be overlooked, leading to missed provision of needed supports. In this study, negative role functioning was rarely reported as an issue of concern in its occurrence or intensity among the parents except during times in which parents performed daily hygiene care for their child. During these moments, parents indicated this was only "sometimes" a factor. In other studies, parents have described the need for them to take on additional roles of medical care provider or become the support system for their partner due to the isolation of the family in efforts to keep their child with CHD healthy (Lee & Rempel, 2011; Meakins, Ray, Hegadoren, Rogers, & Rempel, 2015; Rempel, 2005; Rempel, Blythe, Rogers, & Ravindran, 2012) .

Previous studies have shown parents of medically fragile infants with complex types of CHD have extremely high reports of stress, isolation, and role dysfunction as they provide care during the child's first two staged repairs (Ellinger & Rempel, 2010; Lee & Rempel, 2011; Meakins et al., 2015; Rempel, 2005; Rempel & Harrison, 2007; Rempel, Rogers, Ravindran, & Magill-Evans, 2012). As the child with hypoplastic left heart syndrome ages, parent reports of stress trend more to normative levels, implying that elapsed time since acute events is a factor in parent perspectives of QOL, stress, and family function (Brosig, Mussatto, Kuhn, & Tweddell, 2007). Previous studies (Diffin et al., 2016; Gronning-Dale et al., 2012; Menahem, Poulakis, & Prior, 2008) examining parental stress when having a child with CHD have noted a decline or resolution of stress as their child ages and are consistent with the PinCHeD study where elapsed time was generally positively correlated with better reports in stress levels, QOL, and family functioning.

Study of parental social support has shown inconsistent results. In studies that compared mothers' and fathers' experiences of social support, Lawoko and Soares (2003) reported that mothers had the lowest availability of social support in contrast to fathers, who reported the greatest availability of social interactions. In contrast, Werner (2014) demonstrated that fathers reported having to give more things up and see family members and friends less frequently than mothers. Mothers of a child with CHD tended to have higher stress levels overall whereas fathers reported higher stress levels than normative populations and predictably lower than mothers (Utens et al., 2000). Utens et al. (2000) sampled parents who were awaiting their child's cardiac surgery, whereas the PinCHeD study sampled parents after their child's cardiac surgeries. Both studies concluded mothers have slightly higher stress levels than fathers; however, the PinCHeD study concluded the gender-based differences were not statistically significant, and neither parent group reported acute stress levels. Unfortunately, due to the lack of equal representation of fathers in the literature, it is unclear if this pattern would be noted in larger studies or over the life course of their child with CHD.

Previous studies have recruited parents of a child with specific cardiac defect types such as hypoplastic left heart syndrome, a single ventricle type of pathophysiology, or two-ventricle repaired CHD types such as transposition of the great arteries (TGA) (Brosig, Mussatto, et al., 2007; Caris et al., 2016; Kaugars et al., 2018). The PinCHeD study did not limit the sample of parents based upon a specific CHD defect type. The findings of this study suggested that the type of CHD is not significantly related to parental reports of stress, QOL, and their family's functioning and therefore, cannot be used as a predictor to

identify which parents may need more supportive interventions to mitigate stress, negatively impacted attributes of QOL, and declines in their family's functioning.

Synthesis of Findings among Studies using the PIP

One of the gaps in the literature this study addressed was the vast number of instruments used to measure parental perspectives, making it difficult to make comparisons among the findings. Similar to the PinCHeD study, recent publications have reported parental perceptions when having a CHD using the PIP as a measure for parental stress (Bishop et al., 2019; Caris et al., 2016; Kaugars et al., 2018). The next section focuses on the similarities and differences of the PinCHeD study in comparison to these studies.

Methodologies Used

The recent studies (Bishop et al., 2019; Caris et al., 2016; Kaugars et al., 2018) discussed here, similar to the PinCHeD study, all used a cross-sectional, descriptive design approach and the PIP (Streisand, Braniecki, Tercyak, & Kazak, 2001) as an instrument to measure parental stress. There are numerous, distinct differences in the methodologies used in these studies in comparison to the PinCHeD study. These methodology differences may contribute to the inconsistent results in stress, QOL, and family functioning when comparing the PinCHeD results to recent research studies. These differences are discussed next.

Recruitment and selective sampling. The Caris et al. (2016) study used anonymous email distribution lists to solicit parents of a child with HLHS to participate in their study. Kaugars et al. (2018) used data from the parental forms that were sent to parents whose child with CHD was scheduled to receive a neurodevelopmental assessment. Bishop et al. (2019) recruited their sample from parents who attended their child's outpatient cardiology

appointment. The PinCHeD study mailed recruitment letters to parents who met eligibility requirements and mailed surveys to parents who agreed to participate.

Selective sampling based on the CHD type is not representative of the general population of children with CHD and their families yet is frequently performed within CHD literature. Caris et al. (2016) selectively sampled to include only parents of a child with hypoplastic left heart syndrome. The Kaugars et al. (2018) study selectively sampled to capture parents of a child with CHD and an identified developmental difference (as evidenced by receiving a referral for psychological services). Although the PinCHeD study did not selectively sample based on diagnosis of a developmental difference, nearly 75% of children in the PinCHeD received early intervention services to treat a developmental difference.

The PinCHeD study purposively selected for parent dyads for equal, gender-based subgroups to determine if mothers and fathers responded differently when asked about their experiences and perceptions of their own stress, QOL, and their family's functioning. Participants in the Caris et al. (2016) study were over 86% female, Caucasian, and college educated. Kaugars et al. (2018) collected parent demographics; however, they did not report percentages of mothers and fathers who participated nor gender-based differences among parent reports when discussing findings. The Bishop et al. (2019) sample was largely comprised of mothers (91.3%).

Data collection and instrument selection. Data collection methods and instrument selection also varied among the studies. The PinCHeD study used only a demographics survey and two measures to assess parental stress, QOL, and family functioning, whereas Bishop et al. (2019), Caris et al. (2016), and Kaugars et al. (2018) used a demographics

survey and three measures for their data collection. Caris et al. (2016) used internet technology for distribution and collection of web-based surveys: the Pediatric Quality of Life Inventory (PedsQL), Parenting Stress Index-Short Form (PSI-SF) (Abidin, 1995), and the Pediatric Inventory for Parents (PIP). The PedsQL (Varni, Burwinkle, Seid, & Skarr, 2003) is a parent proxy tool used to measure the parent's perspective of their child's quality of life is different than the PedsQL-FIM (Varni, Sherman, Burwinkle, Dickinson, & Dixon, 2004) which is a parent self-report of the impact the child's condition has on the parents and family. Kaugars et al. (2018) compiled data from parent forms previously collected as part of their child's neurodevelopmental appointment planning: a demographics survey, the Parenting Stress Index-short form (PSI-SF) or Parenting Stress Index, 4th edition (PSI-4) (Abidin, 1995), the PedsQL-FIM (Varni et al., 2004), and the PIP (Streisand et al., 2001). Bishop et al.'s (2019) study used a demographic survey, the Pittsburgh Sleep Quality Index, a measure of psychological symptoms (Brief Symptom Index-18), and the Pediatric Inventory for Parents (PIP). It is unclear if the choice to use different instruments to measure similar parental outcomes impacts the overall results of each study; however, the use of different instruments to measure similar parental outcomes makes it difficult to compare results across studies.

Description of child's CHD type and severity. Description or ranking of CHD types or severity were approached differently among all recent studies. Caris et al. (2016) included only parents of a child with hypoplastic left heart syndrome. Kaugars et al. (2018) delineated CHD type/severity by whether the child had cardiac anatomy consisting of a single ventricle versus two ventricles and split the findings based on categorization for sub-group comparisons. For the Bishop et al. (2019) study, the child's pediatric cardiologist

rated the child's CHD type/severity as "simple," "moderate," or "complex." The PinCHeD study used the empirically derived STAT scoring system (O'Brien et al., 2009) as a method to identify CHD severity. The use of different ranking or categorization of CHD severity may create challenges when attempting to compare results across studies, as the severity of CHD types may be interpreted differently.

Use of theoretical framework. The use of a theoretical framework is not consistent among the studies discussed in this section. Caris et al. (2016) nor Kaugars et al. (2018) included discussion of a chosen theoretical model that guided their research. For their framework, Bishop et al. (2019) used an integrated model derived from the transactional model of stress and coping (Lazarus & Folkman, 1984) and the theory of allostasis and allostatic load (McEwen, 1998; McEwen & Stellar, 1993; Sterling & Eyer, 1988). The selection of the double ABCX theory as a framework is a strength for this study in the manner in which it drove instrument selection for the capturing of variables that influence family functioning and adaptation. The double ABCX also assisted with determining the quantitative analyses that would be used to explore the relationships between the variables.

Stress among Parents of a Child with CHD

The lack of clinically significant levels of stress among this sample is consistent with the findings of other recent outcome studies on parents of a child with CHD (Caris et al., 2016; Kaugars et al., 2018). For example, Caris et al. (2016) studied parents of a child with hypoplastic left heart syndrome (HLHS), and the Kaugars et al. (2018) sample was comprised of parents with identified concerns for their child with CHD warranting a referral for follow-up with a pediatric psychologist (nearly 60% of children had single ventricle pathophysiology). The PinCHeD sample was comprised of 25% parents of a child with

single ventricle pathophysiology or highest severity score of five. Correlations between PedsQL-FIM and PIP aggregate scales in the PinCHeD study were similar to those reported in the Kaugars et al. (2018) study, even though the child's CHD types were collectively not as severe. Caris et al. (2016) looked specifically at parents of children with HLHS, and, although Caris's mean values were higher in each PIP scaled score than in this study, the findings were similar in that neither study demonstrated clinically significant levels of parental stress. The lack of acute scores among parent reports across the PinCHeD, Caris et al. (2016), and Kaugars et al. (2018) studies could be attributed to the development of effective coping skills and strong support systems, and the acquisition of resources.

Stress and developmental differences. The Kaugars et al. (2018) study used selective sampling with the recruitment of eligible parents based on the known presence of psychological or developmental differences in their child with severe forms of CHD. The PinCHeD study showed parents of a child with CHD who had received early interventions services (indicating developmental delays or differences had been identified in the child under three years of age) reported significant levels of anxiety and concern over their child's future or how other family members were affected by the medical condition of the child with CHD. These findings indicate that the child's developmental differences may contribute to higher levels of parental psychological distress and interventions to improve the child's developmental outcomes may also improve the psychological distress parents endorse regarding their child's outcomes and care.

Communication and stress. The parents in the PinCHeD study reported no clinically significant difficulty or stresses related to communication with care providers. Although relatively higher, but not clinically significant, Caris et al. (2016) reported similar

findings with their focused sample of parents of an infant with HLHS. The concept of communication is not often discussed in the literature, yet research demonstrates better parent-staff communication is associated with lower parent stress, while communication between family members improves family cohesiveness and functioning and teaches children problem solving (American Psychological Association, 2019; Hasanpour, Alavi, Azizi, Als, & Armanian, 2017).

Emotional dysfunction and stress. The PinCHeD study found mothers typically report higher emotional dysfunction and more worry than fathers and implies that the time elapsed since their child's cardiac surgery may be a factor in the intensity and frequency that stress and emotional distress are experienced. In this study, all parents of a child with CHD reported higher scores compared to other measured scales when asked specifically about their worry for their child's future and is similar to findings of previous studies (Bevilacqua et al., 2013; Utens et al., 2000).

Effects of time and stress. Previous studies examining parents of a child with CHD and stress have noted a decline or resolution of stress as their child ages. This supports the findings within the PinCHeD study that elapsed time is generally positively correlated with better reports in stress, QOL, and family functioning. As time passes, the child's care management needs become fewer; hence, the stress this causes for parents decreases. The findings regarding the effects of elapsed time within the PinCHeD study are consistent with the evolution of reported stress, QOL, and family functioning among parents of infants with severe CHD types such as hypoplastic left heart syndrome, a severe type of CHD that requires a three-staged surgical palliation course (Brosig, Mussatto, et al., 2007)).

QOL and Family Functioning among Parents of a Child with CHD

Both the Caris (2016) study and the PinCHeD study reported correlations suggesting that high QOL was associated with lower stress levels and higher levels of family functioning. Approximately one-third of parents in the Kaugars et al. (2018) study reported "at-risk" scores in at least one summary scale of the PedsQL-FIM and no statistically significant differences between the single versus two-ventricle cardiac anatomy subgroups.

The parents in the PinCHeD study did not feel isolated or experience a lack of support. This contrasts with findings of earlier research in which parents of a child with CHD reported high levels of loneliness, isolation, and decreased support (Diffin et al., 2016; Doherty et al., 2009; Levert et al., 2016). Parents in the current study did not report difficulty in finding time or feeling up to participating in social activities. This demonstrates a level of bonadaptation in which the participating parents may have learned to take opportunities to meet life values or needs such as leisure activities that foster social activities, positive emotions, and the development of new skills and interests (Brajša-Žganec et al., 2011). Although social expectations generally place mothers as the care provider for the children in the family, the father's role and self-identification as the family's breadwinner may be overlooked, leading to missed opportunities to provide needed supports. In this study, negative role functioning was rarely reported as an issue of concern in its occurrence or intensity among the parents, except during times in which parents performed daily hygiene care for their child. During these moments, parents indicated this was only "sometimes" a factor. In other studies, parents have described the need for them to take on additional roles of medical care provider or become the support system for their partner due to the isolation of the family in efforts to keep their child with CHD healthy (Lee & Rempel, 2011; Meakins et al., 2015; Rempel, 2005; Rempel, Blythe, et al., 2012).

Strengths and Limitations of the Study

Several strengths and limitations have been identified to assist with guiding future research studies. A strength of this study was the equal representation of gender-based parent groups. Equal representation of mothers and fathers allows for the full picture of family experiences and assists in the conceptualization of family functioning. Equal representation of parent genders strengthens the ability to identify differences in the types of support services which may be offered specific to the needs of mothers and fathers.

Another strength of this study is the sampling of parental pairs from same homes to ensure potentially compounding variables (such as, for example, socioeconomic status, number of family members in the home) were decreased or avoided. Sampling parents from same households strengthens the results by decreasing the number of variables that may influence the parent reports. For instance, a mother and father from the same household would likely share the same financial situation; therefore, analyses between the parents would have stronger internal validity by removing alternative explanations, such as different financial situations, for the findings. Sampling parent dyads from same households also strengthens the PinCHeD study by allowing for a framework of similar life experiences and shared supports in the gender-based sub-groups relative to the medical care and health trajectory of each parent dyad's shared child with CHD.

The representation of parents of children with diverse CHD types and comorbidities is another strength of the PinCHeD study, as it allows for greater generalizability to the national population of parents of a child with CHD. There are numerous forms of CHD ranging in severity and often present with co-morbidities (Miller, Riehle-Colarusso, Alverson, Frias, & Correa, 2011). Previous studies have often selectively sampled based on

capturing perspectives of parents of children with the most severe forms of CHD and omitting families of children with less severe types of CHD (Brosig, Mussatto, et al., 2007; Lee & Rempel, 2011).

There are several limitations to the PinCHeD study, the first of which is the homogeneity of the sample. The majority of the parent pairs were middle-class, educated, heterosexual, and Caucasian. Although efforts were made to ensure all communications and instruments were available in Spanish, no Spanish-speaking families agreed to participate. It is a challenge to determine how the PinCHeD study compares to the national population of parents of a child with CHD due to the selective sampling, small sample sizes, and cross-sectional designs predominantly used in the previous research.

The small sample size was another limitation of this study. The a priori study sample size to be fully powered was not achieved. A study with inadequate statistical power has a reduced likelihood of detecting a true effect and increases the probability of making a type 2 error, also known as a "false negative." Simply put, a type 2 error is when the researcher fails to observe a difference when there is one (Button et al., 2013).

Implications of PinCHeD Study

The PinCHeD study was a pilot study that explored the stress, QOL, and family functioning in parents of a child with CHD in the Central Midwest region of the United States and demonstrated feasibility as well as challenges to be addressed in future research. Derived implications from the results of the PinCHeD study can be used to guide practice, theoretical framework selection, policy initiatives, and research methodology.

Practice Implications

The findings of the PinCHeD study implied CHD type or severity was not an accurate predictor of adverse parental perceptions the severity of the child's CHD and should not be used as a method to identify which parents or families are at greatest risk for adverse perceptions of their stress, QOL, and family functioning. This implication contrasts with previous studies, which demonstrated severity of the cardiac defect type should be considered during and beyond the acute phase of medical of care due to the recognition of the child's reduced QOL and its adverse influence on the parental QOL and their support sources (Denniss, Sholler, Costa, Winlaw, & Kasparian, 2019; Verrall et al., 2019). The PinCHeD study demonstrated positive relationships between perceptions of stress, QOL, and family functioning and indicated that changes in one of those areas may influence how other areas are perceived.

The PinCHeD study was purposeful in its sampling to ensure equal representation of fathers and mothers for analysis of differences in areas of psychological and functional impact, since few studies with equal samples have been performed (Brosig, Whitstone, Frommelt, Frisbee, & Leuthner, 2007; Diffin et al., 2016; Doherty et al., 2009). In an interview for the American Heart Association (2017), Dr. Sarah Woolf-King shared there was not a clear understanding of why mothers of a child with CHD disproportionately experienced post-traumatic stress disorder; however, Dr. Woolf-King's (2017) systematic review demonstrated a key reason for this finding: the unequal and under-representation of fathers in related research.

Theoretical Implications

The findings of the PinCHeD study demonstrated the application of the double ABCX theory to guide research design and instrument selection was a logical choice due to the theory's ability to represent the many diverse characteristics and relationships of parental perceptions and family functioning when having a child with CHD. The PIP (Streisand et al., 2001), PedsQL-FIM (Varni et al., 2004), and specific demographic data (such as household income) measured numerous aspects captured in the factors represented in the double ABCX model as well as the relationships between them. The PinCHeD study demonstrated strong relationships between measured factors within the double ABCX theory and indicate that the placement of support interventions in one area of QOL may also improve perceptions in stress and family functioning. The PinCHeD study now adds additional support for goodness of fit when using the double ABCX theory in the application of research focused on perspectives of family members of a child with CHD.

Policy Implications

The implications regarding policy as demonstrated in the PinCHeD study are related to the equal representation of fathers in this study. The PinCHeD study demonstrated fathers do report elevated stress levels, poorer QOL, and poorer family functioning when having a child with CHD and may benefit from policy initiatives that support the ability of the father to participate more actively in the raising of their child with CHD and eliminate potential stressors related to role-functioning and financial strain.

Research Implications

One research implication of the PinCHeD study is the importance of assessing the perceptions of fathers of a child with CHD due to their uniqueness in comparison to those of

mothers. In the PinCHeD study, mothers and fathers demonstrated differences in their perceptions of specific aspects of their stress, QOL, and family functioning. Efforts to understand the unique perspectives of all parents of a child with CHD equally are not typically found within the literature, as mothers are more prevalently represented. The PinCHeD study demonstrated the importance of father-inclusive research when studying families of children with CHD.

Recommendations for Future Research

The PinCHeD study was a pilot study that yielded several recommendations for consideration in future studies in the areas of practice, theory, research, and policy.

Future Recommendations Related to Practice

The close relationships found in this cross-sectional view between parental perceptions of stress, QOL, and family functioning drives a practice recommendation to perform routine, clinical assessments of perceptions of the stress, QOL, and family functioning of all parents of a child with CHD. The performance of routine clinical assessments may assist in the determination of causes and intensity of perceived stress or areas of the parents' QOL or family's functioning that is most adversely affected at specific times in the life course of their child with CHD. Routine, clinical assessments would also assist in determining the effectiveness of interventions and supports that are provided to family members in need.

Future Recommendations Related to Theory

No consistently-used theoretical model that guides hypothesis building or research design exists among the CHD population. The PinCHeD study used the double ABCX theory as framework guiding instrument and statistical analysis using a cross-sectional

design approach where data was gathered after the child with CHD had experienced their surgical repair and a period of time had passed for the family to adjust post-surgery. Selection of a theory that encompasses or considers the influence of time on functioning or adaption is key to goodness of fit. The double ABCX theory is longitudinal in nature and supports the performance of cohort studies that directly examine changes in relationships among CHD family members over a period of time with or without interventional influence. Future cohort studies could be interventional in design with consistent and logical application of the double ABCX theory. Using the double ABCX theory as a unifying theoretical model would assist in construct identification when attempting to synthesize findings across multiple studies.

Coping is a factor within the double ABCX that has not been directly measured in the research on parents of a child with CHD. Lazarus and Folkman (1984) described coping as the cognitive and behavioral changes made in effort to continuously manage internal and/or external demands that are stressful or exceeding a person's resources. The inclusion of a tool to measure coping would add to the overall goodness of fit to the double ABCX theory by representing the coping factor and could be used to demonstrate effectiveness of interventions and supports the family receives.

Future Recommendations Related to Research

The first recommendation bridges the areas of theoretical considerations and future research recommendation and includes a measure that evaluates parental coping to improve goodness of fit when using the double ABCX theory to frame research. Another research-related recommendation also involves instrument selection and the ability to adapt the instrument for use in web-based distribution and submission by participants. Caris et al.

(2016) used web-based self-reports and were able to recruit much larger samples using social network sites such as Facebook and listservs. Using web-based recruitment and data collection methods may pose challenges in ensuring equal representation of parent genders; however, a large-sized participating sample similar to the Caris et al. (2016) study is difficult to achieve in single-site studies.

Another recommendation related to instrument selection is the inclusion of a tool to measure marital satisfaction among married or cohabitating parents. Understanding the dynamics between spouses may be helpful in identifying strengths or weaknesses specific to the relationship between the parents and can assist with determination of the need of marriage-supportive therapies or interventions. Descriptive studies that explore family dynamics, such as relationships between spouses, partners, or siblings, and spillover effects of stress among family members would improve the understanding of role transitions, relationship changes, and magnitude/type of support needs for all family members of a child with CHD (Lavelle, Wittenberg, Lamarand, & Prosser, 2014).

In regard to sampling, efforts should be made to find creative and innovative methods to improve diversity and inclusivity among participants. Although the PinCHeD study was heterogeneous in terms of the CHD types represented, the parent sample lacked diversity. The majority of current research on parents of a child with CHD predominantly focuses on middle-class, Caucasian mothers. Recruitment of a heterogeneous parent sample will improve the generalizability of future studies. Attention to diversity in sampling should be a priority to also ensure multicultural and multigenerational family systems are represented to better understand key points of vulnerability for culturally-sensitive care at interventions (Katz et al., 2018). Other parent populations, such as same-sex couples and

grandparents raising children, have unique perspectives when raising a child with CHD, and researchers should consider research methodology such as multi-site collaborative or case studies to ensure these parents are also represented in the literature.

The PinCHeD study could be repeated and achieve a more heterogeneous sample by recruiting single parents and describing their unique perspectives in comparison to married or co-habitating parents. Single parents may report higher stress levels due to the lack of a supporting partner to share the stressors and burdens having a child with CHD poses. Addressing the unique experiences of single parents of a child with CHD would also contribute to the gap in literature related to single parents of children with chronic illness (Brown et al., 2008).

In studies that examined sibling perspectives among populations with chronic conditions such as diabetes or cancer, siblings frequently endorsed poorer QOL (Lavigne & Ryan, 1979; Woodgate, Edwards, Ripat, Rempel, & Johnson, 2016). Research addressing perspectives of siblings or grandparents of children with CHD is emerging but remains scant (Caris et al., 2018; Ravindran & Rempel, 2011; Redshaw & Wilson, 2012). Previous literature that includes siblings of a child with CHD was frequently neurodevelopmental outcome-related, and heart-healthy siblings served as a control group (McCusker, Armstrong, Mullen, Doherty, & Casey, 2013). Studies that examined QOL reports among CHD siblings demonstrated adjustment problems and more behavioral and internalizing problems than siblings of children with other chronic health conditions (Caris et al., 2018; Havermans, Croock, Vercruysse, Goethals, & Diest, 2015). More research needs to be performed to better evaluate the impact of having a brother or sister with CHD on their siblings' psychosocial outcomes.

Parents of children with chronic health conditions/CHD generally report a decrease in stress and increase in QOL as time passes (Brosig et al., 2013; Diffin et al., 2016; Menahem et al., 2008). The variance in length of time between the child's surgery dates and survey completion by parents would be an influential factor in adaptation that could be assessed in a longitudinal study.

Future Recommendations Related to Policy

The lack of father-inclusive research impacts the development of father-supportive policies. Father-supportive policies are not being initiated as quickly as economic shifts for parents and families are occurring. Slow, progressive changes to U.S. policy are happening that promote and support father involvement for their children and families (National Conference of State Legislatures, 2016). When compared to other developed countries, the United States has a lack of nationwide leave policies for parents and is the only country in the Organisation for Economic Co-Operation and Development (OECD) that does not mandate paid maternal leave at the federal level. Some of these countries even offer parental and "homecare" leave (for either parent) (OECD, 2016). Of the 23 OECD countries with paid parental leave, usage by fathers remains low, and incentive programs are being explored by numerous countries to improve rates of usage by men (2016).

Two areas have been studied more among mothers than among fathers: the specific impact of the parent on child development and the impact of a child's health condition on the parent (Jackson et al., 2015). Many researchers determine this to be due to the considerably less amount of time fathers spend interacting with and caring for their children (Borklund & Jordan, 2013). Although most research supports this determination, there are reports that indicate some fathers in developed countries spend as much or more time with

their children than mothers (Clutton-Brock, 1991; Eibl-Eibefeldt, 1989; Whiting & Whiting, 1975).

After the original publication of the American Academy of Pediatrics' (Coleman, Garfield, & Committee on Psychosocial Aspects of Child and Family Health, 2004) clinical report on the father's role, an upswing in attention and research on fathers' roles in the care and development of their children occurred. This upswing in attention is attributed to the increased work in the areas of academic studies, policy initiatives, and socioeconomic forces (Yogman et al., 2016). Only a few states have recently adopted policy developments that go beyond the federal Family Medical Leave Act (FMLA), include paid family leave laws, and take a new look at adoption leave for fathers (National Conference of State Legislatures, 2016). A call to action for advocacy by health care providers working with families of a child with CHD on behalf of fathers who desire to play an active role in the care and development of their child with CHD is a warranted, admirable endeavor. Development of policies that are supported by research and reflective of the culture shifts and economic trends can go far in the acceptance and promotion of the "new, nurturing, co-parenting father" (Sarkadi et al., 2008). Nursing research that is father-inclusive can bolster advocacy efforts and assist with father-supportive policy initiatives.

Conclusion

Medical and surgical advancements have led to the survival of children with even the most severe types of CHD. To promote outcomes for children with CHD, an emerging focus has been placed on understanding and improving the perceptions of their parents and functioning within their families. This study explored the perceptions of parental stress, QOL, and family functioning when having a child with CHD. It reports findings that

mothers and fathers endorse areas of impact differently and not as acutely as reported in previous studies. Time may be a contributing influence on the intensity of parental perceptions of stress, QOL, and family functioning. Feasibility for continued research was established. Recognition of adjustments to the methodology has been appreciated for future studies. The findings of this study may be useful to parents, educators, healthcare providers, and social workers who work with families of a child with CHD.

APPENDIX A

INSTRUMENTS USED

PEDIATRIC INVENTORY FOR PARENTS

Below is a list of difficult events which parents of children who have (or have had) a serious illness sometimes face. Please read each event carefully, and circle HOW OFTEN the event has occurred for you <u>in the past 7 days</u>, using the 5 point scale below. Afterwards, please rate how DIFFICULT it was/or generally is for you, also using the 5 point scale. <u>Please complete both columns for each item.</u>

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often	HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely
1. Difficulty sleeping	1 2 3 4 5	1 2 3 4 5
2. Arguing with family member(s)	1 2 3 4 5	1 2 3 4 5
3. Bringing my child to the clinic or hospital	1 2 3 4 5	1 2 3 4 5
4. Learning upsetting news	1 2 3 4 5	1 2 3 4 5
5. Being unable to go to work/job	1 2 3 4 5	1 2 3 4 5
6. Seeing my child's mood change quickly	1 2 3 4 5	1 2 3 4 5
7. Speaking with doctor	1 2 3 4 5	1 2 3 4 5
8. Watching my child have trouble eating	1 2 3 4 5	1 2 3 4 5
9. Waiting for my child's test results	1 2 3 4 5	1 2 3 4 5
10. Having money/financial troubles	1 2 3 4 5	1 2 3 4 5

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often					HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely				
11. Trying not to think about my family's difficulties	1	2	3	4	5	1	2	3	4	5
12. Feeling confused about medical information	1	2	3	4	5	1	2	3	4	5
13. Being with my child during medical procedures	1	2	3	4	5	1	2	3	4	5
14. Knowing my child is hurting or in pain	1	2	3	4	5	1	2	3	4	5
15. Trying to attend to the needs of other family members	1	2	3	4	5	1	2	3	4	5
16. Seeing my child sad or scared	1	2	3	4	5	1	2	3	4	5
17. Talking with the nurse	1	2	3	4	5	1	2	3	4	5
18. Making decisions about medical care or medicines	1	2	3	4	5	1	2	3	4	5
19. Thinking about my child being isolated from others	1	2	3	4	5	1	2	3	4	5
20. Being far away from family and/or friends	1	2	3	4	5	1	2	3	4	5
21. Feeling numb inside	1	2	3	4	5	1	2	3	4	5
22. Disagreeing with a member of the health care team	1	2	3	4	5	1	2	3	4	5
23. Helping my child with his/her hygiene needs	1	2	3	4	5	1	2	3	4	5
24. Worrying about the long term impact of the illness	1	2	3	4	5	1	2	3	4	5
25. Having little time to take care of my own needs	1	2	3	4	5	1	2	3	4	5

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often					HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely				
26. Feeling helpless over my child's condition	1	2	3	4	5	1	2	3	4	5
27. Feeling misunderstood by family/friends as to the severity of my child's illness	1	2	3	4	5	1	2	3	4	5
28. Handling changes in my child's daily medical routines	1	2	3	4	5	1	2	3	4	5
29. Feeling uncertain about the future	1	2	3	4	5	1	2	3	4	5
30. Being in the hospital over weekends/holidays	1	2	3	4	5	1	2	3	4	5
31. Thinking about other children who have been seriously ill	1	2	3	4	5	1	2	3	4	5
32. Speaking with my child about his/her illness ..	1	2	3	4	5	1	2	3	4	5
33. Helping my child with medical procedures (e.g. giving shots, swallowing medicine, changing dressing).....	1	2	3	4	5	1	2	3	4	5
34. Having my heart beat fast, sweating, or feeling tingly.....	1	2	3	4	5	1	2	3	4	5
35. Feeling uncertain about disciplining my child	1	2	3	4	5	1	2	3	4	5
36. Feeling scared that my child could get very sick or die.....	1	2	3	4	5	1	2	3	4	5
37. Speaking with family members about my child's illness	1	2	3	4	5	1	2	3	4	5
38. Watching my child during medical visits/procedures	1	2	3	4	5	1	2	3	4	5
39. Missing important events in the lives of other family members	1	2	3	4	5	1	2	3	4	5

EVENT	HOW OFTEN? 1=Never, 2=Rarely, 3=Sometimes, 4=Often, 5=Very often	HOW DIFFICULT? 1=Not at all, 2=A little, 3=Somewhat, 4=Very much, 5=Extremely
40. Worrying about how friends and relatives interact with my child	1 2 3 4 5	1 2 3 4 5
41. Noticing a change in my relationship with my partner	1 2 3 4 5	1 2 3 4 5
42. Spending a great deal of time in unfamiliar settings	1 2 3 4 5	1 2 3 4 5

ID#_____

Date:_____

PedsQL™

Family Impact Module

Version 2.0

PARENT REPORT

DIRECTIONS

Families of children sometimes have special concerns or difficulties because of the child's health. On the following page is a list of things that might be a problem for **you**. Please tell us **how much of a problem** each one has been for **you** during the **past ONE month** by circling:

0 if it is **never** a problem
1 if it is **almost never** a problem
2 if it is **sometimes** a problem
3 if it is **often** a problem
4 if it is **almost always** a problem

There are no right or wrong answers.
If you do not understand a question, please ask for help.

*In the past **ONE month**, as a result of your child's health, how much of a problem have **you** had with…*

PHYSICAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel tired during the day	0	1	2	3	4
2. I feel tired when I wake up in the morning	0	1	2	3	4
3. I feel too tired to do the things I like to do	0	1	2	3	4
4. I get headaches	0	1	2	3	4
5. I feel physically weak	0	1	2	3	4
6. I feel sick to my stomach	0	1	2	3	4

EMOTIONAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel anxious	0	1	2	3	4
2. I feel sad	0	1	2	3	4
3. I feel angry	0	1	2	3	4
4. I feel frustrated	0	1	2	3	4
5. I feel helpless or hopeless	0	1	2	3	4

SOCIAL FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel isolated from others	0	1	2	3	4
2. I have trouble getting support from others	0	1	2	3	4
3. It is hard to find time for social activities	0	1	2	3	4
4. I do not have enough energy for social activities	0	1	2	3	4

COGNITIVE FUNCTIONING *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. It is hard for me to keep my attention on things	0	1	2	3	4
2. It is hard for me to remember what people tell me	0	1	2	3	4
3. It is hard for me to remember what I just heard	0	1	2	3	4
4. It is hard for me to think quickly	0	1	2	3	4
5. I have trouble remembering what I was just thinking	0	1	2	3	4

COMMUNICATION *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I feel that others do not understand my family's situation	0	1	2	3	4
2. It is hard for me to talk about my child's health with others	0	1	2	3	4
3. It is hard for me to tell doctors and nurses how I feel	0	1	2	3	4

*In the past **ONE month**, as a result of your child's health, how much of a problem have **you** had with…*

WORRY *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. I worry about whether or not my child's medical treatments are working	0	1	2	3	4
2. I worry about the side effects of my child's medications/medical treatments	0	1	2	3	4
3. I worry about how others will react to my child's condition	0	1	2	3	4
4. I worry about how my child's illness is affecting other family members	0	1	2	3	4
5. I worry about my child's future	0	1	2	3	4

DIRECTIONS

Below is a list of things that might be a problem for **your family**. Please tell us **how much of a problem** each one has been for **your family** during the **past ONE month.**

*In the past **ONE month**, as a result of your child's health, how much of a problem has **your family** had with…*

DAILY ACTIVITIES *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Family activities taking more time and effort	0	1	2	3	4
2. Difficulty finding time to finish household tasks	0	1	2	3	4
3. Feeling too tired to finish household tasks	0	1	2	3	4

FAMILY RELATIONSHIPS *(problems with…)*	Never	Almost Never	Some-times	Often	Almost Always
1. Lack of communication between family members	0	1	2	3	4
2. Conflicts between family members	0	1	2	3	4
3. Difficulty making decisions together as a family	0	1	2	3	4
4. Difficulty solving family problems together	0	1	2	3	4
5. Stress or tension between family members	0	1	2	3	4

Parent Demographics Form

***Internal Use Only*-Record ID: _____**

Parent Name (Last, First): _____

Date of Birth: ____ /_____/_____

What sex were you assigned at birth?: ☐ Male ☐ Female

What is your current gender identity?:

☐ Male ☐ Female ☐ Choose not to disclose

☐ Additional gender category, please specify: _____

Of the child with congenital heart disease, I am the:

☐ Biological Mother	☐ Step/ Bonus Mother	☐ Adoptive Mother Father	☐ Legal Guardian/ Foster Parent
☐ Biological Father	☐ Step/ Bonus Father	☐ Adoptive Father	☐ Other, specify: _____

Race (mark all that apply):	**Ethnicity (choose one):**
☐ White ☐ Black or African American ☐ American Indian and Alaska Native ☐ Canadian and Latin American Indian ☐ Asian ☐ Native Hawaiian And Other Pacific Islander ☐ Some other race	☐ Hispanic or Latino origin (of any race) ☐ White alone, not Hispanic or Latino

Your Highest Completed Level of Education
☐ No schooling completed ☐ Kindergarten ☐ Grade 1 through 11 – Specify grade 1 – 11: _____ ☐ 12th grade NO DIPLOMA

☐ Regular HIGH SCHOOL DIPLOMA ☐ GED or alternative credential ☐ Some college credit, but less than 1 year of college credit ☐ 1 or more years of college credit, no degree ☐ Associate's degree (for example: AA, AS) ☐ Bachelor's degree (for example: BA, BS) ☐ Master's degree (for example: MA, MS, MEng, MEd, MSW, MBA) ☐ Professional degree beyond a bachelor's degree (for example: MD, DDS, DVM, LLB, JD) ☐ Doctorate degree (for example: PhD, EdD)
<u>Annual Household Income</u> **Include: Salary & wages, rental income, Supplemental Security Income (SSI), public assistance or welfare payments, retirement, survivor, or disability pensions, and all other income:**
☐Less than $10,000 ☐15,000 to $24,999 ☐$25,000 to $34,999 ☐$35,000 to $49,999 ☐$50,000 to $74,999 ☐$75,000 to $99,999 ☐$100,000 to $149,999 ☐$150,000 to $199,999 ☐$200,000 or greater

Occupation:_____

Household Size (the number of all people who occupy your housing unit, ie. apartment, mobile home, group of rooms, or a sing room that is occupied as separate living quarters)?: _____

Total number of children under the age of 18 living in the household: _____

Number of grandparents of the child with CHD living in the household: _____

What is your current relationship status?
☐ Married- Legal or Common law- Spouse/Partner Present in household (includes same-sex couples) ☐ Married- Legal or Common law- Spouse/Partner <u>NOT</u> Present in household (includes same-sex couples) ☐ Unmarried, Spouse/Partner-Present in household (includes same-sex couples) ☐ Unmarried, Spouse/Partner-<u>NOT</u> Present in household (includes same-sex couples) ☐ Widow/Widower
How many years have you been in this relationship? _____

APPENDIX B

LETTERS OF SUPPORT AND PERMISSION

April 27, 2017

Mary (Becky) Gregory, MSN RN CNOR
PhD Student
School of Nursing and Health Studies
University of Missouri-Kansas City
Kansas City, Missouri 64080

Miss Becky Gregory:

I am pleased to offer my support for your STTI grant proposal "A descriptive exploration of quality of life and perceived impact on family functioning in parents of a child with a congenital heart defect: The PinCHeD study." As your PhD Committee Chair, I fully support this study.

I understand that members of the Cardiac Neurodevelopmental and THRIVE Programs collaborate frequently to promote best outcomes and supportive interventions for your patients and their families. Identifying CHD parent perceptions regarding their own quality of life and family function will assist in understanding which approaches for interventions and services are most frequently needed. Again, I offer my full support for this much needed work.

I wish you success in obtaining funding for this project and look forward to continued collaboration with you in the future.

Sincerely,

Patricia Kelly, PhD, MPH, APRN
Professor, School of Nursing and Health Studies
University of Missouri-Kansas City
Phone: 816-235-2617
kellypj@umkc.edu

Children's Mercy
KANSAS CITY

2401 Gillham Road
Kansas City, Missouri 64108
(816) 234-3000

April 28, 2017

Mary (Becky) Gregory, MSN RN CNOR
PhD Student
School of Nursing and Health Studies
University of Missouri-Kansas City
Kansas City, Missouri 64080

Dear Ms. Gregory:

I am pleased to offer my full support for your STTI grant proposal "A descriptive exploration of quality of life and perceived impact on family functioning in parents of a child with a congenital heart defect: The PinCHeD study." Members of the Cardiac Neurodevelopmental and THRIVE Programs collaborate frequently to promote best outcomes and supportive interventions for our patients and their families. Identifying CHD parent perceptions regarding their own quality of life and family function will assist in understanding which approaches for interventions are most frequently needed. I am excited about the possibility of this project as information obtained from it may allow us to provide more targeted and evidence-based supportive services. With 15+ years of experience in both clinical care and research in the fields of neuropsychology and pediatric psychology, I believe that I am well-positioned to support you in this important work.

I wish you success in obtaining funding for this project and look forward to continued collaboration with you.

Sincerely,

Elizabeth Willen, Ph.D. | Psychologist
Division of Developmental and Behavioral Sciences – Psychology Section
Ward Family Heart Center
Children's Mercy Kansas City
P: (816) 234-3674 ext. 56258
E: ejwillen@cmh.edu
2401 Gillham Road | Kansas City, MO 64108

Children's Mercy
KANSAS CITY

2401 Gillham Road
Kansas City, Missouri 64108
(816) 234-3000

April 23, 2018

Mary (Becky) Gregory, PhD(c) RN CNOR
PhD student
School of Nursing and Health Studies
University of Missouri-Kansas City
Kansas City, Missouri 64080

Miss Becky Gregory:

As Director of the Ward Family Heart Center and Medical Director of the Cardiac Neurodevelopmental (CND) Program, I fully support your study, "Quality of life and perceived impact on family functioning in parents of a child with congenital heart disease: The PinCHeD study."

Members of the Cardiac Neurodevelopmental and THRIVE Programs collaborate frequently to promote best outcomes and supportive interventions for our patients and their families. Identifying CHD parent perceptions regarding their own quality of life and family function will assist in understanding which approaches for interventions and services are most frequently needed. This study will serve as a strong foundational work for a trajectory of research describing this patient population.

I wish you success in obtaining funding for this project and look forward to continued collaboration with you in the future.

Sincerely,

James E. O'Brien, Jr., MD, FACS
Co-Director, Ward Family Heart Center
Chief, Section of Cardiothoracic Surgery
Medical Director, Cardiac Neurodevelopmental Program
Jerry Smith Chair in Pediatrics
Children's Mercy Hospital
Associate Professor of Surgery
University of Missouri - Kansas City School of Medicine
2401 Gillham Road
Kansas City, MO 64108
Phone: (816) 234-3580
Fax: (816) 302-9946
jobrien@cmh.edu

From: STREISAND, RANDI < RSTREIS@childrensnational.org>
Sent: Tuesday, March 28, 2017 11:33 AM
To: Gregory, Becky, R
Cc: Gregory, Mary R. (UMKC-Student)
Subject: RE: Request to use PIP in dissertation project
Attachments: PIP Materials 2017.doc

*** This message was sent to you from an External Source. Please do not open untrusted links or attachments. ***

Hi Becky-
Thanks for your interest in the PIP. Sure – you are welcome to use the measure in your dissertation work.

See attached.

Best of luck-
Randi Streisand

Randi Streisand, Ph.D., CDE
Professor of Psychology & Behavioral Health, and Pediatrics
Center for Translational Science- Team Leader for Behavioral& Community Research
Director of Psychology Research
Children's National Health System
111 Michigan Ave., NW
Washington, D.C. 20010
202-476-2730
We stand for children.
rstreis@childrensnational.org
www.ChildrensNational.org

From: Gregory, Becky, R [mailto:mrgregory@cmh.edu]
Sent: Monday, March 27, 2017 1:46 PM
To: STREISAND, RANDI
Cc: Gregory, Mary R. (UMKC-Student)
Subject: Request to use PIP in dissertation project

Hello Dr. Randi Streisand,

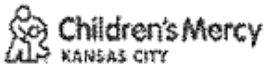
I am currently a PhD-Nursing student at the University of Missouri , Kansas City. I am writing to request use of the Pediatric Inventory for Parents (PIP) as an instrument to measure parenting stress variables in my dissertation project. My population of interest is parents of a child with a congenital heart defect and my dissertation work is a descriptive study of parental QOL and perceived family functioning when having a child with a congenital heart defect. I have enjoyed the research I have found in other chronic pediatric health conditions that use this instrument and have not found many studies in the pediatric cardiology discipline that have used the PIP as an instrument. I would like to address this gap in the literature by integrating the PIP into my research.

Please let me know if there any requirements for use of this instrument. If there are none or minimal, would you please consider granting me permission to use this instrument in my research, beginning with my

dissertation project, and provide a copy of the PIP instrument, administration and scoring instructions, and other associated guides? Of course, all academic acknowledgements and citations will be made.

Feel free to contact me if you have any questions or would like additional information about my research project and use of the PIP.
Thanks for all you do,

Becky Gregory, MSN RN CNOR | Program Coordinator
Cardiac Neurodevelopmental Program
Children's Mercy Kansas City
P: (816) 983-6019 || F: (816) 802-9946
E: mrgregory@cmh.edu | W: childrensmercy.org
2401 Gillham Road | Kansas City MO 64108



SPECIAL TERMS

These User License Agreement Special Terms ("Special Terms") are issued between Mapi Research Trust ("MRT") and Mary Gregory ("User").

These Special Terms are in addition to any and all previous Special Terms under the User License Agreement General Terms.

These Special Terms include the terms and conditions of the User License Agreement General Terms, which are hereby incorporated by this reference as though the same was set forth in its entirety and shall be effective as of the Special Terms Effective Date set forth herein.

All capitalized terms which are not defined herein shall have the same meanings as set forth in the User License Agreement General Terms.

These Special Terms, including all attachments and the User License Agreement General Terms contain the entire understanding of the Parties with respect to the subject matter herein and supersedes all previous agreements and undertakings with respect thereto. If the terms and conditions of these Special Terms or any attachment conflict with the terms and conditions of the User License Agreement General Terms, the terms and conditions of the User License Agreement General Terms will control, unless these Special Terms specifically acknowledge the conflict and expressly states that the conflicting term or provision found in these Special Terms control for these Special Terms only. These Special Terms may be modified only by written agreement signed by the Parties.

1. **User information**

User name	Mary Gregory
Category of User	Student
User address	2401 Gillham rd Kansas City 64108 Missouri United States of America
User VAT number	
User email	mrgregory@cmh.edu
User phone	816-983-6019
Billing Address	2401 Gillham rd Kansas City 64108 Missouri United States of America

2. **General information**

Effective Date	Date of acceptance of these Special Terms by the User
Expiration Date ("Term")	Upon completion of the Stated Purpose
Name of User's contact in charge of the request	Mary Gregory

3. **Identification of the COA**

Name of the COA	PedsQL™ - Pediatric Quality of Life Inventory™
Author	Varni JW
Copyright Holder	Varni James W, PhD
Copyright notice	Copyright © 1998 JW Varni, Ph.D. All rights reserved
Bibliographic reference	List of references for each PedsQL™ module
Modules/versions needed	PedsQL™ Family Impact module

4. **Context of use of the COA**

The User undertakes to use the COA solely in the context of the Stated Purpose as defined hereafter.

4.1 Stated Purpose

Clinical Practice

Type of use*	Individual practice
Planned Term*	Start: 11/2018; End: 09/2019
Number of screened patients	219
Number of sites	1
Number of submissions of the COA for each patient	2
Mode of administration*	Paper
If electronic administration, please indicate mode of data collection	
Use of IT Company (e-vendor)	No

4.2 Country and languages

MRT grants the License to use the COA on the following countries and in the languages indicated in the table below:

Version/Module	Language	For use in the following country

PedsQL™ Family Impact module	English	the USA
PedsQL™ Family Impact module	Spanish	the USA

The User understands that the countries indicated above are provided for information purposes. The User may use the COA in other countries than the ones indicated above.

5. **Specific requirements for the COA**

- The Copyright Holder of the COA has granted ICON LS exclusive rights to translate the COA in the context of commercial studies or any project funded by for-profit entities. ICON LS is the only organization authorized to perform linguistic validation/translation work on the COA.

- In case the User wants to translate the COA in an academic context, the User shall send the back translations to the Copyright Holder for approval

- In case the User wants to use an e-Version of the COA, the User shall send the Screenshots of the original version of the COA to the Copyright Holder through MRT for approval. The Copyright Holder may request consulting fees for this review

- In case the User wants to use an e-Version of the COA, the User shall send the Screenshots of the translations of the COA to ICON LS for approval.

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VITA

Mary Rebecca “Becky” Gregory was born July 22, 1972 in Adrian, Michigan. She was educated at Bowling Green High School in Bowling Green, Ohio. She started her family and then began her educational pursuits in Nursing. She was awarded a Bachelor of Science in Nursing in 2003 from Missouri Western State College in Saint Joseph, Missouri. She continued her educational and professional pursuits and earned her Master’s degree in Nursing with a focus in Leadership and Executive Administration from the University of Missouri-Kansas City in 2009.

In addition to the pursuit to obtain a Ph.D. in Nursing, Ms. Gregory has spent over 16 years in various nursing roles including as a direct care peri-operative nurse, nurse consultant, and nursing manager and leader. She has worked at Mosaic Hospital, St. Luke’s Hospital, and Children’s Mercy Hospital in critical care positions including the genitourinary/gynecology, general-vascular, cardio-thoracic, and outpatient peri-operative specialties. She has held leadership and consultant positions at Truman Medical Center, Aesculap, Inc., and most recently, Children’s Mercy Hospital as the Cardiac Neurodevelopmental Program Coordinator/ Manager.

Ms. Gregory entered the University of Missouri-Kansas City with a research focus on understanding and improving the experiences of families of children with congenital heart disease. Ms. Gregory plans to continue working in the nursing community to serve as a leader and educator to nurses and to improve the lives of families touched by chronic health care conditions through continued research and education of community and hospital-based care providers.