CAREGIVER KNOWLEDGE OF RISK FACTORS ASSOCIATED WITH COMPLEX CONGENITAL HEART DISEASE AND QUALITY OF LIFE OUTCOMES

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Congenital heart disease is the most common birth defect globally, affecting both children and their families. Twenty-five percent of children experiencing a CHD birth defect are diagnosed with complex CHD (cCHD), signifying critical heart dysfunction requiring one or more open-heart surgeries during the first year of life. With medical advances, cCHD survival rates have almost tripled in the last three decades. This has resulted in an increase in the number of morbidities associated with cCHD, which is drastically impacting the need to support quality of life outcomes for a child with cCHD and their family. The two most prevalent unaddressed risks for quality of life outcomes in the cCHD population are child and caregiver mental health and child's neurodevelopmental disabilities. The present study sought to address the relationship between caregiver knowledge of cCHD developmental challenges (i.e., outcomes related to neurodevelopmental disabilities, mental health, and provider quality-care approach) and children's and caregiver's quality of life outcomes, inclusive of academic functioning ability of children with cCHD, children with cCHD and their caregivers' mental health functioning, and the overall satisfaction with the nature of the healthcare provider of the child with cCHD. A total sample size of $N = 46$ participants were included in the current study. Results that caregivers' knowledge of cCHD risks to quality of life outcomes explained a much greater percentage of the variance in caregiver satisfaction with healthcare providers ($R^2 = 0.350$, $p < 0.001$) compared to number of surgical interventions ($R^2 = 0.058$, $p = 0.047$). Clinical implications and implementation for use of a holistic, integrated approach are strengthened by the study findings.

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CAREGIVER KNOWLEDGE OF RISK FACTORS ASSOCIATED WITH COMPLEX CONGENITAL HEART DISEASE AND QUALITY OF LIFE OUTCOMES

Introduction

Congenital heart disease (CHD) is a malformation within the structure of the heart that occurs during fetal development (McCusker & Casey, 2016). CHDs are the most prevalent type of birth defect, occurring in approximately 40,000 births annually in the United States, remaining the leading cause of birth defect mortality (CDC). Twenty-five percent of CHD children are diagnosed with critical CHD (cCHD), indicating critical heart dysfunction requiring one or more cardiac surgical interventions for life sustainability within the first year of life (Garcia et al., 2016; Ruggiero et al., 2018).

Over the last several decades medical, surgical, and technological advancements have dramatically improved complex cardiac interventions and long-term treatment for the cCHD population. These advances have significantly increased the life expectancy for children with cCHD (Triedman & Newburger, 2016). With this change came the necessity for healthcare teams to shift from a predominantly medical focus to inclusion of understanding and managing developmental risk factors that may impact children with cCHD and their families' quality of life (QOL; Bellinger & Newburger, 2010). For example, the healthcare team, family, and children's educators need information to form a clear understanding of potential developmental and physical challenges for a child with cCHD as they reach school age (Wang et al., 2014).

Currently, however, the majority of cCHD research and interventions remain centered on the medical components of care for children with cCHD, and less on developmental factors associated with QOL for the child with cCHD and family (Marino et al., 2016). The absence of research addressing these core child- and family-related issues leaves a major gap in cCHD

healthcare practice and research. The absence of this shared information can impact families' knowledge of medical, mental, and social function (Daily et al., 2016). The importance of a broader focus is highlighted by research indicating a connection between children's medical problems and high risks of psychopathology and mental health disorders in their caregivers and siblings (O'Connor et al., 2016; Wang et al., 2014).

The current study explores what factors may be related to positive QOL outcomes for children with cCHD and their families. Specifically, we look at how caregivers' knowledge of their school-aged children's specific developmental needs impacts quality of life issues, such as satisfaction with the information shared by their children's healthcare providers, their level of mental health as care providers, and their children's academic performance. [Please note, caregiver is defined in the current study as a parent, family member, or any other individual who are primarily responsible for caretaking and medical decisions of a child with cCHD.]

Risk Factors Experienced by Children with cCHD and Their Families

Level of Caregiver Knowledge

In parenting a child with a chronic illness, caregivers benefit from having general information about children's development, as well as information specific to risk factors and experiences related to the child's illness. The importance of being knowledgeable about children's developmental needs and potential risks experienced within cCHD families are reflected in literature on parental self-efficacy (Barlow & Ellard, 2004; Patterson, 2003; Jackson et al., 2016) and parental perception of illness (Garcia et al., 2016; Wei et al., 2015; Wang et al., 2014).

A caregiver's lack of knowledge and awareness regarding a cCHD diagnosis, with feelings of uncertainty of how to care for and meet the needs of their child, evokes feelings of

helplessness, fear, and high chronic stress within the family environment. Psychoeducation, knowledge provided to caregivers addressing needs and uncertainties, has shown to be the most significant intervention for cCHD adaptive family functioning, a major key component of quality of life (QOL). Results indicate a statistically significant increase in caregiver self-efficacy, specifically empowerment and competence ability in caring for the child with cCHD, with decreased feelings of uncertainty (Barlow & Ellard, 2004; Patterson, 2003; Jackson et al., 2016).

The child with cCHD's home environment characteristics, specifically caregiver wellbeing and caregiver perception of the child's cCHD illness, are indicated as the strongest predictors for overall child developmental outcomes and family QOL, regardless of cCHD illness type or severity (Garcia et al., 2016; Wei et al., 2015; Wang et al., 2014). Caregivers that are knowledge about their child's development are involved and attuned to their child's needs (Lubkin, & Larsen, 2013). Garcia et al. (2016) highlights the vital role and impact of parent wellbeing on a children's development, and the significance of modeling healthy coping strategies. Parental perception of the cCHD illness has shown to be positively correlated to family level QOL (Garcia et al., 2016; Wei et al., 2015; Wang et al., 2014). Providing caregivers with knowledge of developmental stages and outcomes, as well as providing information about risk factors related to different parenting practices, has a positive impact on child outcomes and family functioning (Jackson et al., 2016; Lantin-Hermoso et al., 2017).

Environmental Influence

An area of QOL that has been largely overlooked within cCHD clinical research pertains to nonclinical contributing factors, environmental factors, and the potential impact on the child with cCHD and family. The family system and home environment are essential for healthy functioning and adaptability (Patterson, 2003). Environmental family system factors can have a

substantial impact on the child with cCHD and family's QOL, influencing the child with cCHD's overall life trajectory and the family functioning ability/adaptability (Animasahun et al., 2015). Parent education and income level, family health related habits (dietary, exercise, routine health screenings), cCHD health outcomes, and family collective view/behavior towards the cCHD illness are all environmental family factors associated with QOL for families with children who have cCHD (Dionysia, 2010; Cassedy et al., 2013). Parent educational level and income are positively associated with child with cCHD health outcomes and academic achievement.

Thus, with children with cCHD's longer survival rates identifying and creating mechanisms to address the developmental risk factors children with cCHD may face in relation to cognitive development, social skills, and physical well being. Based on available research, children's psychosocial and neurodevelopmental delays (Marino et al., 2012; Wei et al., 2015) and families' and children's quality of mental health (Woolf-King et al., 2018) are the most prevalent unaddressed developmental risk factors faced by children with cCHD and their families (Janssens et al., 2016; Brosig et al., 2017). Each of these risk factors and current research related to these risk factors are addressed below.

Caregivers' Mental Health

The most common mental health diagnoses experienced by caregivers, as well as their child with cCHD, are anxiety, depression, and post-traumatic stress disorder (Shillingford et al., 2008). A recent cross-sectional study in the Netherlands found over 80% of cCHD caregivers reported feeling clinically significant symptoms of trauma; 50% reported feeling elevated symptoms of depression and/or anxiety; and up to 80% reported feeling severe psychological distress (Helfricht, et al., 2009). Caregivers' described feelings of poor mental health as being intermittent, comparing the experience to constantly being on a roller coaster ride, uncertainty

and constant emotional ups and downs (Wei et al., 2016).

Mental health has a bidirectional association to family functioning ability. Maladaptive family functioning occurs when the family of the child with cCHD is unable to adapt or re-adapt to the needed family restructuring required for caring for a chronically ill child with cCHD (Garcia et al., 2016; Patterson, 2003). Mental health plays a key role in the caregiver-child relationship and healthy family functioning (Woolf-King et al., 2018).

The importance of caregiver mental health for QOL and child outcomes is evidenced in that caregiver mental health is positively associated with the child with cCHD's mental health functioning and adaptability – directly influencing the prevalence of internal/external behavior problems in children with cCHD (Kolaitis et al., 2017;Wei et al., 2015). Caregiver mental health is a stronger predictor of QOL outcomes for children with cCHD than is that of the type of long-term illness experienced by the child and/or illness severity, including surgical factors (Woolf-King et al., 2018). Despite this rate of mental health challenges, a recent study 70% of cCHD caregivers reported that their mental health and that of their children were not being addressed or acknowledged by healthcare providers (Hutchinson & Middlemiss, 2019).

Children's Neurodevelopmental and Psychosocial Development

Children with cCHD are at high risk of neurodevelopmental disabilities and psychosocial delays that can impact their executive functioning and academic success (Marino et al., 2012; Wernovsky & Light, 2016). The incidences of neurodevelopmental sequelae and psychosocial comorbidity prevalence in children with cCHD is very high (Brosig et al., 2017; Marino er al., 2016; Marino et al., 2012). Over 50% of children with cCHD have a neurodevelopmental disability and children with cCHD are three times more likely to need educational intervention services than same age peers without a heart defect (Riehle-Colarusso et al., 2015).

Neurodevelopmental disabilities in children with cCHD are attributed to a multitude of potential contributing risk factors, one major contributing factor being the heart disease itself. In addition, the number of surgical interventions increases health risks, including neurodevelopmental disabilities (Garcia et al., 2016).

Biopsychological Healthcare Approach

As risk of morbidity and comorbidity associated with cCHD increases, developing care-team approaches designed to increase knowledge, resources, and psychoeducational interventions that introduce and address associated risks is increasingly important. Needed across healthcare environments (e.g., hospital-based, community-based) are advances that help medical practitioners shift from bio-medical, isolated approaches to utilizing biopsychosocial approaches (Utens et al., 2018). A biopsychosocial approach is a holistic, interconnected approach focused around the relationships of the biological, psychological, personal, family, and community (McDaniel et al., 2013). This type of approach helps families with children with cCHD address existing risks and increase healthy family functioning and QOL (Marino et al., 2012).

Toward this goal, the American Heart Association (AHA) released the first scientific statement clearly identifying the high risk and prevalence of neurodevelopmental and psychosocial impairments within the cCHD population (Marino et al., 2010). The formal statement provided cardiac medical providers with a step-by-step guideline for screening and treatment management to be implemented with all cCHD patients, integrated as standard care practice (Garcia et al., 2016; Marino et al., 2012). One of the main objectives for the formal statement release was to signify how early intervention was vital for decreasing long-term developmental delays (i.e., significantly lower academic and socio-emotional functioning) associated with unaddressed or undiagnosed issues (Ringle & Wernovsky, 2016).

Formal American Academy of Pediatrics (AAP) guidelines for cCHD medical providers (AAP, 2017) had a similar focus. The AAP guidelines proposed that healthcare providers adopt a patient-centered standard of care across care providers. This care approach was defined as collaborated care and patient-centered placing emphasis on the caregivers as part of the medical treatment team. With this approach, there is a focus on information sharing, transparency, and holistic decision making between medical providers and caretakers (Lantin-Hermoso et al., 2017).

The patient-centered care approach is suggested as a step that may positively impact the current challenges families and medical providers face in relation to sharing information. From a similar focus, the Association for European Pediatric and Congenital Cardiology (AEPC) recently proposed a new medical care model, one in which the care and needs of the children with cCHD and their family are addressed holistically and from a multidisciplinary approach (Utens et al., 2018). The family-centered psychosocial care model is multilevel, with care provided to the child and family when the child is receiving inpatient and outpatient care, as well as across the lifespan (Utens et al., 2018). The development of guidelines for treatment of cCHD highlight the prevalence of unaddressed risks and the need for implementing interventions and providing available resources supporting cCHD families.

Despite these guidelines that included recommendations for family inclusion as part of the care team, the potential risks faced by a child with cCHD are widely unknown to many cCHD caregivers (Marino, 2010;). Further, these risks are often novel to many educators and clinical providers (Wei et al., 2015; Marino et al., 2016; Marino et al., 2010). According to Wei et al. (2017), cCHD caregivers' knowledge may be limited by what information is accessible and shared by their child's healthcare providers.

Current Study

The present study explores the importance of caregiver knowledge of developmental risk factors for children with cCHD in relation to child and family outcomes. The three research questions addressed are:

RQ1. Is caregiver knowledge of risk factors experienced by children with cCHD (including caregiver knowledge of neurodevelopmental disabilities, mental health risks for the caregiver and/or child with cCHD, and healthcare provider quality care [as measured by the Leuven Knowledge Questionnaire –Congenital Heart Disease (LKQ-CHD)]) significantly positively correlated to caregivers' satisfaction with their healthcare provider (treatment and care approach),[as measured by the PedsQL Healthcare Satisfaction Form (HSF).

RQ2. Is caregiver knowledge of cCHD risk factors (including caregiver knowledge of neurodevelopmental disabilities, mental health risks for the caregiver and/or child with cCHD, and healthcare provider quality care [as measured by the Leuven Knowledge Questionnaire – Congenital Heart Disease (LKQ-CHD)]) a stronger predictor variable than demographic and medical covariates (caregiver data included age, education level, and annual income; child with cCHD data included number of surgical interventions and current age) of both the caregivers' satisfaction with the healthcare provider [PedsQL (HSF)] and their child's QOL outcomes [as rated on the (PCQLI)]?

RQ3. Is caregiver knowledge of cCHD risks, including caregiver knowledge of neurodevelopmental disabilities, mental health for the caregiver and/or child with cCHD, and healthcare provider quality care [as measured by the Leuven Knowledge Questionnaire – Congenital Heart Disease (LKQ-CHD)]) significantly positively correlated with the academic functioning of children with cCHD [as measured by the CBCL] and/or their own level of mental

health [as measured by the PCL-5 and PQH-9]?

Methods

Participants

Caregivers (N = 46) were recruited using purposeful sampling techniques. Eligibility requirements included that caregivers be the primary care provider of a child diagnosed with cCHD who 5 years was or older and had attended school outside the home. Exclusion criteria included the caregivers or caregivers partner having a diagnosis of cCHD or congenital heart disease, caregiver currently being part of a cCHD-related clinical trial study, or the focus child having been diagnosed with a gene syndrome or other serious debilitating comorbid medical illness.

Data Collection

Purposive sampling was conducted through online, closed cCHD caregiver support group websites [e.g., Congenital Heart Disease Community and Awareness (N = 8,894 members); and Mended Little Hearts of Fort Worth resource website (N = 450)]. A flyer with information about the current study was posted on both nationally based online support websites. The flyer and research project were approved by the University of North Texas Institutional Review Board (IRB).

To assure confidentiality of participants responses, all participants were assigned a random ID number when they began the study. All data were stored and filed according to the IRB guidelines. Caregivers who received the flyer online could click on a link that would bring them to the introductory page of the study. This page provided study details and those caregivers signed a consent form. Participants did not receive compensation for completing the survey.

Procedures

The procedure of conducting caregiver participation online allowed data collection from a more heterogeneous, less location bound sample than would in-person participation. There was a direct link available on the flyer to redirect participants to the main information study page on University-sponsored Qualtrics software program. The main information page provided more in-depth content than that which was listed on the flyer. Participants were able to contact the researchers by email for any unanswered questions or concerns. Information was available to participants for support if needed and resource information within the CHD community, and national mental health website and hotline phone number.

After the consent forms were completed, participants filled out an online questionnaire to assess and confirm that the participant met the study proposal criteria. Participants meeting the study criteria clicked a tab taking them to the first page of the study surveys and when prompted, continued to the first survey question. Caregivers who were not eligible were sent to a page thanking them for considering participation.

The first survey questions addressed caregiver family demographic information and cCHD medical history information. Family demographic information included caregiver age, sex, education level, annual income, and race and current age of the child with cCHD. Medical history information collected pertained to the cCHD diagnoses and the number of surgical interventions to date. Four additional survey questions completed at this step focused on two major deficits: mental health and neurodevelopmental risks. Responses provided participants' reports regarding whether mental health and neurodevelopmental risks were discussed by their children's healthcare team (see Appendix A).

The next survey completed by participants was the LKQ-CHD questionnaire (see Moons

et al., 2001) measuring caregiver knowledge of cCHD and risk related to children's developmental outcomes as related to neurodevelopmental disabilities, mental health related issues, and healthcare provider quality care) The additional surveys completed included the PCL-5 (Weathers et al., 2003) and the PHQ-9 (Kroenke et al., 2001), which assessed caregiver's mental health symptoms (trauma and depression) and mental functioning; school competency portion of the CBCL (Achenbach & Rescorla, 2917); which assessed academic functioning; the PedsQL (HSF; Lewis et al., 1986), assessing provider quality of care provided to patients and families and provider quality-care approach type used with cCHD caregivers. The final survey completed was the PCQLI questionnaire (Uzark et al., 2003), assessing QOL for children and adolescents with cCHD using a parent proxy form.

Data Collection

Data collection was ended following a 4-week period of no new participants in response to three reposting of the invitation. This study limitation necessitated a change in planned analyses in that the total sample size would not allow completion of a one-way multivariate analysis of variance (MANOVA). The number of participants was not sufficient to meet the assumptions to conduct an MANOVA with all criterion variables at once (Cohen, 1988). Descriptive data collected addressed responses from four descriptive questions on caregiver's experience with their cCHD's healthcare provider and knowledge sharing of mental health and neurodevelopmental risks/preventative resource information. These analyses are presented in Appendix A and are not presented as part of this study.

Instruments

Leuven Knowledge Questionnaire for Congenital Heart Disease (LKQ-CHD)

The Leuven Knowledge Questionnaire for Congenital Heart Disease (LKQ-CHD; see Moons et al., 2001) is a research instrument developed to measure knowledge as related to CHD (Moons et al., 2001). The LKQ-CHD revision is necessary to address caregiver's knowledge of their child with cCHD within the pediatric population, whereas the current self-reporting version measures adults cCHD knowledge of their own diagnosis and disease (permission was obtained from Moons et al., 2001 to revise subdomains for the current study and use of the instrument was granted for data collection).

The LKQ-CHD assesses knowledge of neurodevelopmental and psychosocial delays, the knowledge of mental health risks and the knowledge of provider treatment care approach models and quality of care. The LKQ-CH is scored dichotomously, 1 = *correct*, 0 = *incorrect*, and measures one variable per domain. The questionnaire is scored as a total knowledge score, based on the number of questions that were answered correctly and the total number of questions available on the knowledge questionnaire. Scoring requires the total score to be calculated, then divided by the total number of eligible answers and multiplied by 100 (Yang et al., 2012; Moons et al., 2001). Thus, the LKQ-CHD score can range from 0 to 100 where lower scores indicate less knowledge of neurodevelopmental and psychosocial delays. Total scores lower than 50% of the total possible score of 100 are viewed as having a low knowledge level and 80% or greater as having a high level of knowledge (Moons et al., 2001). Previous research reports psychometric properties for the LKQ-CHD as indicating high validity I-CVI = .78, Kappa = .896, Liu et al., 2016). In the present study, internal consistency was high ($\alpha = .78$).

PedsQL Healthcare Satisfaction Form (HSF)

The PedsQL (HSF; Lewis et al., 1986) assesses caregivers' perception of the healthcare team quality of care as provided to their child and the family. The self-report questionnaire

consists of items that include information on inclusion of family, partnership-coordination of care, communication and trust, emotional needs, and overall satisfaction. Response descriptors range from *never* (0) to *always* (3) on a 4-point Likert scale. The sum of the scores for each item represented a total symptom score. The scale does not require reverse coding. Lower scores indicate more satisfaction. The PedsQL (HSF) indicates strong validity, internal consistency (α = .78) and test–retest reliability (r =.79), both above acceptable levels (Li et al., 2013). Cronbach's alpha in this study was .95.

Pediatric Cardiac Quality of Life Inventory

The Pediatric Cardiac QL (QL) Inventory (PCQLI; Uzark et al., 2003) is a disease-specific questionnaire that assesses the QOL outcomes for children and adolescents with congenital heart disease. The PCQLI originates from the health-related QOL questionnaire, and currently is in use at major hospitals throughout the United States (Marino et al., 2010; Marino et al., 2016). The PCQLI is the currently the only cardiac disease specific measure to be generalizable within countries outside of the Unites States (Marino et al., 2010).

The PCQLI comprises 21 items and two subscales: Disease Impact (physical functioning) and Psychosocial Impact (psychological, social, and emotional functioning). Item 21 of the survey is ambiguous, the emotional functioning subscale question, and not included as part of the overall QOL total score (Marino et al., 2010). Therefore, Item 21 was omitted from the computation of the PCQLI score for this study as necessary. The remaining 20 items were used to calculate the score.

The parent form of the PCQLI consists of response scores ranging from 1 (*strongly agree*) to 5 (*strongly disagree*) on a 5-point Likert scale (Marino et al., 2016). The total score is as the sum of responses to the 20 survey statements. Thus, the total score can take on a range of

values from 20 to 100, where lower scores indicate a lower QOL. Parent proxy form internal consistency (α = .71 to .90) and test retest reliability (.70) are both above acceptable levels. In this study, Cronbach's alpha was .93.

Child Behavior Checklist (CBCL)

The CBCL school competence scale (CBLC/6-19; Achenbach & Rescorla, 2017) assessed academic functioning in cCHD elementary aged children. The CBCL has been used extensively with children in the cCHD population and includes scores for competency scales, syndrome scales, and DSM-5-oriented scales applicable to a wide age range in children (Carlson et al., 2014; McCusker et al., 2012). For the current study, the CBCL school competency items assess caregivers' view of their children's academic functioning and educational delays or need for support.

Items are scored on a 3-point Likert scale, with lower scores indicating lower school competency abilities. Results were available in raw data and *t*-score formats, with standard scoring available for comparison across sex and multicultural based norms. Estimated time needed to complete this assessment is approximately 10 minutes. Questions pertain to a child's typical grades/academic achievement and the need for educational support/school intervention. Psychometric properties are strong for internal validity (α = .73 to .91) and test-retest reliability (r = .73) for the CBCL (Achenbach & Rescorla, 2017). In the present study, Cronbach's alpha was .92.

Post-Traumatic Stress Disorder Checklist for DSM-5 (PCL-5): Mental Health

The PCL-5 (Weathers et al., 2013) is a self-report questionnaire that assesses *DSM-5* symptoms of PTSD. Self-report rating scale descriptors range from *not at all* (0) to *extremely* (4) on a 5-point Likert scale. The sum of the scores for each item represented a total symptom score.

The scale does not require reverse coding. Completion of the PCL-5 requires approximately 5 to 10 minutes to complete. The PCL-5 is a psychometrically sound DSM-5 assessment, both valid and reliable (Weathers et al., 2013). Internal consistency ($\alpha = .91$), test–retest reliability ($r = .61$), and concurrent, convergent, and discriminant validity ($r = .69$) were at acceptable levels (Ghazali & Chen, 2018). In the present study, Cronbach's alpha was .96.

Patient Health Questionnaire, Depression (PHQ-9)

The PHQ-9 (Kroenke et al., 2001) is a self-report questionnaire assessing depressive symptomology on a 4-point Likert scale, ranging from *not at all* (0) to *everyday* (3). The sum of question responses reference a depressive severity score summary. Specifically, the sum of the scores for each item represented a total symptom score. The scale does not require reverse coding. Higher scores for each scale indicate higher psychopathology symptoms for depression and/or PTSD. The PHQ-9 shows strong validity, internal consistency ($\alpha = .89$), and reliability for testing and measuring depressive symptomology (Kroenke et al., 2001). In the present study, Cronbach's alpha was .94.

Planned Analysis

Initial examination of all demographic information was conducted. Demographic and medical data consisted of caregiver and the child with cCHD information. Caregiver data included age, sex, education level, and annual income; race, number of surgical interventions, and current age were collected on the child with cCHD. After all data was analyzed, sex of the caregiver and race of child were not used in additional analyses throughout study due to homogeneous data.

Pearson's correlation and Spearman's rho correlation analyses were used to answer RQ 1, which examined the relationship between caregiver knowledge of cCHD risk factors (mental

health, neurodevelopmental disabilities, provider quality care), and healthcare provider satisfaction level (criterion variable). Both variables for RQ 1 were on continuous measurement scales. Pearson's correlation analysis assesses for a potential relation or linear trend. Given the relatively small sample size often a Spearman' rho was conducted to look for agreement (Tabachnick & Fidell, 2013; Liu et al., 2016).

For RQ2, bivariate correlations between demographic and medical covariates (caregiver data included age, education level, and annual income; child with cCHD data included number of surgical interventions and current age) and caregiver knowledge of cCHD risk factors, as measured by the LKQ-CHD and the two dependent variables, PCQLI and PedsQL (HSF), were computed. A univariate analysis of variance (ANOVA) examined each component as related to RQ 2 (Tabachnick & Fidell, 2013). Bonferroni adjusted two-sample *t*-tests were performed to determine group statistical significance. The Bonferroni correction controls error rate, or Type 1 error that can commonly occur when conducting multiple analyses on the same dependent variable (Tabachnick & Fidell, 2013). A hierarchical multiple linear regression then assessed any significant findings within the dependent variable and independent variables (both knowledge and demographic factors) that were statistically significant within the ANOVA analyses.

Pearson's correlation and Spearman's rho correlation analyses were both used to address RQ 3. In RQ 3, three subquestions were assessed separately, as each was unrelated to the other. The three subquestions broken down individually addressed (1) the association between caregiver knowledge of cCHD risks and the academic functioning of children with cCHD; (2) the association between caregiver knowledge of cCHD risks caregivers' and caregiver post-traumatic stress disorder; and (3) the association between caregiver knowledge of cCHD risks and caregiver depression.

Results

Demographics

Of the 46 caregivers meeting the inclusion criteria, 36 (78.3%) were between the ages of 26 and 45 years, and 10 (21.7%) were between the ages of 46 and 65 years; all 46 identified as female. Caregivers provided responses in relation to one focus child; 30 of the focus children with cCHD were between 5 and 10 years of age (65.2%) and 16 were 11 years of age or older (34.8%). The highest level of education reported by the 46 caregivers was between high school and Associates degree (*n* = 14, 30.4%) and Bachelors, Masters, Ph.D. (*n* = 32, 69.6%). Participants' household income was between $21,000 and $80,000 (*n* =17, 37.0%), and $81,000 or above (*n* = 29, 63.0%). The number of heart surgeries reported for each child with cCHD ranged from 1 to 7 surgeries with 21 children reported as having 1 to 2 surgeries (45.7%), 25 children having 3 to 7 surgeries (54.3%). All focus children were identified as White.

Table 1

Descriptive Statistics for Six Scales

	N		Mean	SD	Min	Max
	Valid	Missing				
PTSD	46	0	17.089	13.3	0.0	44.0
Depression	46	0	9.33	7.73	0.0	25.0
Satisfaction with HealthCare	46	0	16.70	6.20	4.0	24.0
Academic Functioning	46	0	6.44	3.28	0.0	12.0
Quality of Life	46	0	59.65	16.27	32.0	100.0
Knowledge	46	0	43.04	23.55	0.0	86.7

SD = Standard Deviation

SAT = Satisfaction with Healthcare Providers as measured by the PedsQL-HSF instrument.

COMP = Academic Functioning - School Competency as measured by the CBCL instrument.

QOL = Quality of Life of the Child with CCHD as measured by the PCQOLI instrument.

KNO = Knowledge of Congenital Heart Disease as measured by the LKQ-CHD instrument.

RQ 1

Figure 1 shows a linear relation between the two variables satisfying the linearity assumption. Results indicated no significant outliers. Both variables had a normal distribution based on inspection of histograms of the independent and dependent variables. Pearson's correlation analysis showed there was a statistically significant, positive correlation between the predictor variable, caregiver knowledge, and the criterion variable, caregivers' satisfaction with their healthcare provider (treatment and care approach), $r(44) = 0.59$, $p < 0.001$ with a $R^2 = .35$ effect size. Spearman's rho correlation analysis has similar findings, $r_s = 0.58$, $p < 0.001$.

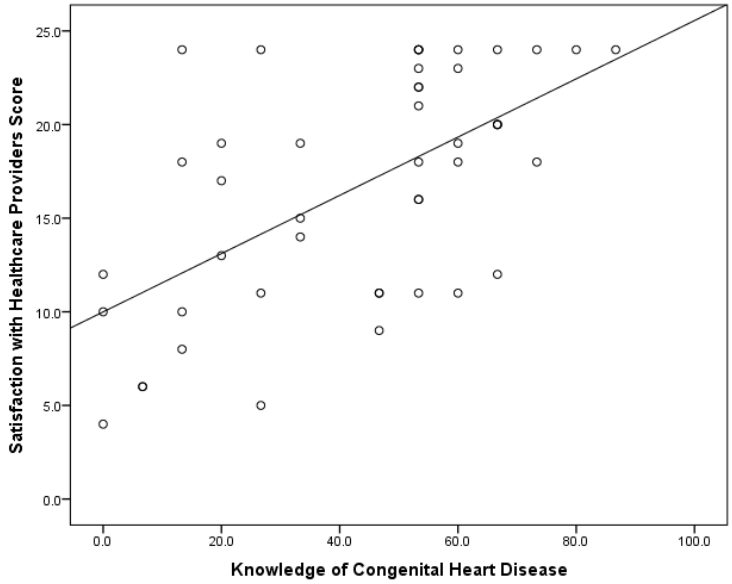

Figure 1. Linear relationship between caregiver knowledge and satisfaction with healthcare provider.

Due to a relatively small sample size, Spearman's rho correlation analysis was performed to compare with the Pearson's correlation findings, since Spearman's rho correlation statistic is unaffected by outliers and non-normal distributions. The Pearson's correlation statistic and the Spearman's rho correlation statistic both identified the same associations findings, indicating the statistically significant results were not likely to have been influenced by the sample size (Tabachnick & Fidell, 2013).

RQ 2

RQ 2 Box plots were created to identify possible existing outliers associated with responses on the caregiver knowledge scale, all demographic variables, and the caregiver satisfaction with their healthcare provider scale. All assumptions to support ANOVA were met. The Shapiro-Wilk Test was used to evaluate normality assumption, and independence, and the Levene's test of equality of variance was used to evaluate the assumption of homogeneity of variance.

Child outcomes as measured by the PCQLI was not statistically significantly related to any of the five demographic variables: caregiver age, education level, annual income, current age and number of surgeries. All partial eta squared effect sizes were below significance (Cohen, 1989). In relation to satisfaction with the healthcare provider, only the independent variables, caregiver knowledge (as reported in RQ 1) and the demographic variable of annual household income were positively statistically significantly associated with satisfaction with their health care provider as shown in Table 3. The remaining four demographic variables were not statistically significantly associated with the dependent variable satisfaction with health care provider as measured by the PedsQL (HSF).

Table 2

Shapiro-Wilk Test to Evaluate the Normality Assumption of the ANOVA for Annual Household Income and Satisfaction with Healthcare Providers

Caregiver Annual Household Income	Shapiro-Wilk		
	Statistic	Df	*p*-value
$21,000 - $50,000	0.93	8	0.54
$51,000 - $90,000	0.91	14	0.12
Greater than $90,000	0.88	24	0.01

There was a statistically significant difference in the average satisfaction with health care provider score among the three income groups, $F(2, 43) = 3.28$; $p = 0.047$; partial $\eta^2 = 0.13$; observed power = 0.593. According to Cohen (1989), small, medium, and large effect sizes for ANOVA are partial $\eta^2 = 0.01$, partial $\eta^2 = 0.059$, and partial $\eta^2 = 0.14$, respectively. Thus, the observed effect size of partial $\eta^2 = 0.13$ is a large effect size.

Table 3

ANOVA Analyses for the Relation between Income and Satisfaction with the Healthcare Provider

	Sum of Squares	Df	Mean Square	F	*p*-value
Between Groups [a, b]	228.531	2	114.265	3.282	0.047
Within Groups	1497.208	43	34.819		
Total	1725.739	45			

a. Partial $\eta^2 = 0.13$

b. Observed power = 59.3%

Table 4

Bonferroni Adjusted Two-Sample t*-Tests for Income and Healthcare Provider Satisfaction*

(I) Caregiver Annual Household Income	(J) Caregiver Annual Household Income	Mean Difference (I-J)	Std. Error	*p*-value
$21,000 - $50,000	$51,000 - $90,000	3.1250	2.6152	0.716
	Greater than $90,000	-1.9583	2.4090	1.000
$51,000 - $90,000	$21,000 - $50,000	-3.1250	2.6152	0.716
	Greater than $90,000	-5.0833	1.9844	0.042
Greater than $90,000	$21,000 - $50,000	1.9583	2.4090	1.000
	$51,000 - $90,000	5.0833	1.9844	0.042

Bonferroni adjusted two-sample *t*-tests analyses was computed to determine which income groups were statistically significant related with the dependent variable healthcare provider satisfaction (Keselman et al., 1998). Table 4 shows the only difference among the

three income groups was in the $51,000 - $90,000 caregiver annual household income group that had a statistically significantly lower average satisfaction with healthcare provider score compared to the income group of $90,000 or above, ($p = 0.042$).

The results for RQ 1 and 2 showed both caregiver knowledge of cCHD risk factors and caregiver household income were statistically significantly associated with caregiver satisfaction with healthcare providers. A hierarchical multiple linear regression analysis was performed (Tabachnick & Fidell, 2013) to examine how caregiver knowledge of cCHD and demographic variables predicted caregiver satisfaction with healthcare providers compared to either cCHD knowledge or any of the demographic variables alone. Prior to conducting the analysis, it was necessary to recode INC into dummy variables, also known as indicator variables (Tabachnick & Fidell, 2013). Specifically, the caregiver annual household income group $51,000 - $90,000 was the referent group. The two dummy variables were coded as: INC1 = 0 if caregiver annual household income was not $21,000 - $50,000, or 1 if caregiver annual household income was $21,000 - $50,000; INC2 = 0 if caregiver annual household income was greater than $90,000.

The independent variable of caregiver knowledge was entered into the model first because it had the strongest association with satisfaction with health care providers ($r(44) = 0.59$, $p < 0.001$). Model 1 shows caregiver knowledge was statistically significantly associated with satisfaction with health care providers ($p < 0.001$) and adding caregiver annual household income to the model did not explain a statistically significant amount of additional variance in satisfaction with health care providers ($p = 0.069$ for INC1, and $p = 0.078$ for INC2).

Considering income was not statistically significantly associated with satisfaction with health care providers after controlling for caregiver knowledge, it was of interest to determine if any of the other four demographic variables of caregiver age, age of child with cCHD, caregiver

education, and number of child's surgeries might explain a statistically significant amount of variance in satisfaction with health care providers when controlling for caregiver knowledge.

Table 5

Model 1: Hierarchical Multiple Linear Regression Analysis Examining Caregiver Knowledge as Related to Income and Satisfaction with Healthcare Provider

Model [a]		Unstand. Coeff.		Stand. Coeff.	T	*p*-value
		B	Std. Error	Beta		
1 [b]	(Constant)	9.998	1.56		6.39	<0.001
	Knowledge of Congenital Heart Disease	0.156	0.03	0.592	4.87	<0.001
2 [c]	(Constant)	7.956	1.80		4.43	<0.001
	Knowledge of Congenital Heart Disease	0.149	0.03	0.568	4.51	<0.001
	INC1	4.067	2.18	0.252	1.86	0.069
	INC2	3.081	1.71	0.251	1.81	0.078

a. Dependent Variable: Satisfaction with Healthcare Providers Score

b. Independent Variable: Knowledge of Congenital Heart Disease

c. Independent Variables: Knowledge of Congenital Heart Disease, and Caregiver Annual Household Income where: the income referent group was $51,000 to $90,000, INC1 = 0 if caregiver annual household income was not $21,000 - $50,000, or 1 if caregiver annual household income was $21,000 - $50,000; INC2 = 0 if caregiver annual household income was not greater than $90,000 or 1 if caregiver annual household income was greater than $90,000.

First, income was removed from the model and caregiver knowledge was retained. Then, the remaining demographic variables, caregiver age, age of child with cCHD, caregiver education, and number of child's surgeries, were dummy coded, using the same method as was necessary for the above Model 1. The dependent variable remained satisfaction with health care provider and again the predictor variable caregiver knowledge was entered into the model first. Then, stepwise model selection was used to determine if any of the dummy coded demographic variables made a statistically significant contribution to the model in terms of explaining variance in satisfaction with health care provider, above and beyond the variance in satisfaction

explained by caregiver knowledge of risks to QOL outcomes.

Model 2 shows that when controlling for caregiver knowledge, only the number of surgical interventions explained a statistically significant amount of additional variance in satisfaction with health care providers above and beyond the variance explained by caregiver knowledge. Specifically, the amount of variance in satisfaction with health care providers explained by caregiver knowledge was $R^2 = 0.350$, the additional amount of variance explained by number of surgeries, after controlling for caregiver knowledge, was 0.058. In total, caregiver knowledge explained $R^{2=}.35$ and number of surgeries explained $R^2=.058$, accounting for 40.8% of the total variance in satisfaction with health care providers scores.

The model indicates that when controlling for the number of surgeries, the average satisfaction with health care providers score increases by 0.17 points for every 1-point increase in the caregiver knowledge score. When controlling for the caregiver knowledge score, the average satisfaction with health care providers score is expected to be 4.2 points larger for caregivers with a child that has had one surgery, compared to caregivers with a child that has had two or more surgeries.

Table 6

Model 2: Hierarchical Multiple Linear Regression Analysis Examining Caregiver Knowledge as Related to Number of Surgical Interventions with Healthcare Provider

	Unstand. Coeff.		Stand. Coeff.	T	*p*-value
	B	Std. Error	Beta		
(Constant)	8.771	1.625		5.399	<0.001
KNO [b]	0.169	0.032	0.644	5.361	<0.001
SURGINT1 [c]	4.194	2.047	0.246	2.049	0.047

a. Dependent Variable: Satisfaction with Healthcare Providers Score

b. Predictor Variable: Knowledge of Congenital Heart Disease, $R^2 = 0.350$

c. Predictor Variable: SURGINT1 (0 = 2 or more surgeries, 1 = 1 surgery), R2 = 0.058

RQ 3

This question addressed whether there was an association between caregiver knowledge of cCHD and either children's academic functioning as measured by the CBCL or caregivers' mental health functioning, i.e., caregiver post-traumatic stress disorder as measured by the PCL-5, and caregiver depression as measured by the PHQ-9. Bivariate comparisons were computed between the independent variable, caregiver knowledge and each of the three dependent variables of academic functioning of school competency, PTSD score, and depression score. Pearson's correlation was used to evaluate the association separately for the independent variable and each of the three dependent variables. All assumptions for Pearson's correlation are meet (i.e., linear relation between the independent and dependent variables, no significant outliers, and a normal distribution, based on inspection of relevant scatter plots).

No statistically significant correlation between caregiver knowledge of risk factors experienced by children with cCHD and children's academic functioning, $r(44) = 0.02$ $p = .91$. The results of the Spearman's rho correlation analysis were in close agreement, $r_s = - 0.01$, $p = 0.97$ indicating sample size did not influence the finding. Pearson's correlation analysis indicated no statistically significant correlation between caregiver knowledge and caregiver PTSD, $r(44) = -0.163$, $p = .28$. The results of the Spearman's rho correlation analysis were in close agreement, $r_s = - 0.048$, $p = 0.75$ indicating sample size did not influence the finding. Pearson's correlation analysis showed no evidence of a statistically significant correlation between caregiver knowledge of cCHD risks and caregiver depression, $r(44) = -0.15$ $p = .32$. The results of the Spearman's rho correlation analysis were in close agreement, $r_s = - 0.072$, $p = 0.63$.

Discussion and Implications

Caregiver Knowledges

This study supported the importance of caregiver knowledge of the potential risk factors experienced by children with cCHD and their caregivers. First, caregivers with greater knowledge of potential risk factors reported being more satisfied with their healthcare provider. This contributes to the existing substantial body of literature indicating the relationship between healthcare providers and caregivers impacts the well-being of both the caregiver and the child with cCHD. Results also indicated parents of children who had experienced a greater number of surgeries were also more likely to have a higher level of satisfaction with their health care providers. This did not account for as much variance in the relation as did level of caregiver knowledge of risks and health care provider satisfaction.

Data on caregiver knowledge was computed, indicating the average level of knowledge in cCHD caregivers is considered relatively low, as measured by the LKQ-CHD (moons et al., 2001). One explanation for the low level of knowledge found in caregivers may be the result of a potentially biased sample. More specifically, members from the same online community group website may share the same ideologies and knowledge level comprehension, ultimately influencing the data and study results.

Implications

Long term cCHD developmental outcomes and children's and families' QOL outcomes are significantly higher when care is provided by a cardiac medical provider that implements a provider-caregiver shared treatment team approach (McDaniel et al 2013). Caregivers' awareness of psychosocial impairments were associated with success in academia peer acceptance, increases early intervention implementation and higher quality parent-child relationship (Marino et al., 2016). A recent study found that over half of cCHD caregivers rate themselves at adequately informed regarding their child with cCHD's long-term care, yet less

than half had objectively sufficient knowledge. Although caregiver knowledge relating to cCHD risks and QOL developmental outcomes are limited, mother's involvement has been identified as an influential factor in identification and early diagnoses of developmental deficiencies, early implementation of interventional methods, and decreases in additional developmental delays (Brosig et al., 2017).

American Academy of Pediatrics (AAP) guidelines for cCHD medical providers highlights the risks cCHD families may encounter. Proposed in the guidelines is a patient-centered approach to adopt as standard care. The collaborative care approach is centered around information sharing, transparency, and holistic decision making between medical providers and caretakers (Lantin-Hermoso et al., 2017). The suggested development of the guidelines for treatment of cCHD highlight the prevalence of unaddressed risks and the need for interventions and available resources to implement within cCHD families.

Mental Health Scores

Mental health issues are often experienced by the whole family with a child diagnosed with cCHD. Mental health is not only detrimental to the caregiver, but the caregiver's mental health is detrimental to the child with cCHD and the overall family functioning ability. Mental health is bidirectional, and debilitating to the whole family. The relationship between mental health and QOL have been well established within prior literature, yet the current study did not find any significant association to mental health or QOL.

Caregiver's mental health was assessed for the current study in two specific areas, PTSD and depression. Although there were no significant findings related to caregiver mental health and QOL or healthcare satisfaction, caregiver's mental health data was evaluated. Results indicate that 46% of the cCHD caregivers scored above the cut off score necessary to meet

standards for clinical diagnosis for PTSD, provisional. Results for measuring cCHD caregiver depression indicated 59% meet criteria for diagnosis of moderately severe to severe clinical depression, 28% meet criteria for diagnosis of mild to moderate clinical depression, and the remaining 13% would not meet criteria for clinical depression. The COVID-19 pandemic may have influenced the level of depression and PTSD symptoms reported by cCHD caregivers.

Implications

The implications for the current study findings for caregivers mental health, specifically PTSD and depression, suggest the 2020 COVID -19 pandemic that was currently active during data collection may have influenced the level of depression and PTSD symptoms reported by participants. The alternative implication is that caregivers with a child diagnosed with cCHD have higher prevalence of PTSD and depression, and those are unrelated or not associated with caregiver knowledge of risks associated with cCHD. In addition, results could potentially be influenced by the participant sample, as stated in the discussion related to the age of the child with cCHD. Lastly, it is possible that the caregivers that participated in the current study are unaware of their low level of knowledge related to cCHD, therefore the absence of risks influence perspective of QOL, thus influencing caregivers mental health. The findings raise the question of how much impact the current pandemic influenced outcome results in the current study and how much variation would occur in the findings if not for the current global pandemic.

Resources for caregiver psychoeducation and family support programs would help with many of the identified concerns. Intervention methods aim to enhance caregiver knowledge, self-efficacy, skills, and deepen caregivers' understanding of the cCHD child's diagnosis, prognosis, and long term quality care (including potential risks/risks as the child ages), and interventional methods/resources for challenges or support needed for optimal outcomes for cCHD risks

(Jackson et al., 2016Mental health and other coping difficulties of caregivers and siblings influence and shape the cCHD child's experience and perspective of living with chronic illness, and would benefit greatly from medical providers integrated, holistic family biopsychosocial approach (McDaniel et al., 2013; Frankle et al., 2003). Assessing the functionality and mental health of the family at pediatric medical visits, in hospital inpatient settings, before surgical procedures, and any other major cCHD high stress situation would be optimal, helping guide cCHD families to lower anxiety, uncertainty, and promote adaptability.

Nonclinical Factors

The nonclinical data collected for the current study included caregiver age, education level, and annual income; children with cCHD data included number of surgical interventions and current age. Data was collected from caregivers of children with cCHD who were age 5 and older.

Number of Surgical Interventions

Parents of children who had experienced a greater number of surgeries were more likely also to have a higher level of satisfaction with their health care providers. This did not account for as much variance in the relation as did caregiver knowledge of risks to their child's QOL outcomes. It may be a connection between number of surgeries and the amount of interaction between a medical team and a parent and family. This may account both for the higher level of knowledge and the satisfaction with the information received. It may be, as well, that children who experience more frequent surgeries are experiencing more severe symptoms with their cCHD. Thereby, a greater amount of conversation is a necessity between parent and the medical team.

Age of Child with cCHD

Age of the child with cCHD was collected to examine the association and potential relationship to the current study outcome variables. No statistically significant relationship was found with QOL, healthcare satisfaction, or academic achievement. Caregivers that participated in the study had children that ranged in age, beginning with five years to 18 years. The age of the child may have impacted the study findings.

Younger children could potentially be associated with lower quality of life as younger children are more likely to be still experiencing surgery and hospitalizations at a much higher frequency than the older children, and are at high risk for difficulties at the beginning of school, neurodevelopmental delays. In addition, caregivers with an older child may have less knowledge of cCHD risks due to their child surviving well as the age of the child progresses and may not have received the same information when their children were younger as caregivers with young children currently are suppose to receive by their healthcare provider regarding cCHD risks.

Implications

Children with cCHD that are both younger in age or older in age may have influenced the study findings for all 5 surveys. Implications suggest younger children could possibly be negatively associated with all five survey outcomes, whereas older children could potentially be positively associated with all five survey outcomes. The current study would have benefited from collecting additional medical data as it relates to the child's current age, such as when the last surgical intervention occurred and within a smaller child age range. Future research to improve the current study findings are addressed in the following section.

Homogeneous White Sample

The race of the child with cCHD was collected during the first stages of the study. All

participants identified their child as White. The lack of diversity in the current study is not uncommon within this area of research. The lack of diversity within literature and current research continues to add to the already existing health disparities.

Prior research has indicated an association that exists within the United States between race and healthcare. Living in low poverty communities and neighborhood deprivation areas decreases the probability that women of color will have health insurance or access to quality healthcare, including prenatal care (Kucik et al., 2014). Migrant women may not seek out prenatal care due to immigration status, and fear of deportation. Hispanic and migrant women have the highest uninsured healthcare status in the United States. Women of color are at higher risk for experiencing risks for pediatric medical services and access to familial community resources due to geographic location of residency and transportation (Castro et al., 2016). The chronic stress and inequality that women of color experience increases the probability of pregnancy and delivery complications, mortality during childbirth, and significant weathering (Collins et al., 2017). A recent qualitative study addressing health disparities in White and Non-White chronically ill patients showed a significant difference in illness knowledge and access. Further highlighting the need to address these disparities.

Health Disparities are defined as differences that exist among specific population groups in the United States in the attainment of full health potential that can be measured by differences in incidence, prevalence, mortality, burden of disease, and other adverse health conditions (NIH, 2014). This reveals differential health outcomes across identity groups and populations. Health disparities can stem from health inequities, systematic differences in the health of groups and communities occupying unequal positions in society that are avoidable and unjust (Graham, 2004). There are major deficits in examining associations between race and ethnicity and cCHD

outcomes, differences in mortality rates, available healthcare (ability to obtain healthcare, quality of healthcare), SES, and family geographic location (Kucik et al., 2014). Race and culture are a vital component of the diagnosis, prevalence, and treatment of the cCHD child and family.

Implications

Examining how these factors are nested within cCHD families and developmental outcomes is vital for healthy family functioning to be implemented within all cCHD families and for developing effective interventions. Previous research indicates that race and ethnicity are predictive of specific cCHD defects and subtypes (Knowles et al., 2017). One must consider the interplay of race, culture, biological, and environmental, influence on the cCHD diagnosis, prognosis, and QOL for the family.

Implications for minority caregivers and cCHD children suggest higher risks of negligence from healthcare providers and access to available resources due to environmental factors (employment schedule, location of cardiologist, transportation, medical insurance, lack of culturally competent healthcare providers). Ultimately resulting in the reinforcement of a dysfunctional cycle of chronic stress within the family system, leaving no clear path for adaptive change (Artiga, et al., 2016).

QOL Outcomes

Both caregiver knowledge and all nonclinical environmental and medical factors were found to have no association to QOL outcomes. This yields the need for a more in-depth exploration, as the current study findings with QOL are at odds with a substantial amount of prior existing research on cCHD QOL outcomes.

It would seem based on the findings with this group of caregivers, that demographic variables, i.e., variance in the family background and parent experience, were not significant

contributors to caregivers' knowledge of risks to children's QOL outcomes—nor their own QOL issues. This is an interesting finding in that it has both positive and potentially negative implications. First, it is possible that further exploration of this finding would indicate that children with cCHD and their families receive similar levels of care and sharing of knowledge with their medical teams. The findings raise the possibility that the nature of the care does not vary based on families' income, caregivers' education level, and that it does not necessarily change over the life course of the child. This may be a reflection of the necessity of medical care for children with this disorder or that those without resources do not have children that live into adulthood, or past the age of attending primary school (beyond high school). However, this may mean as well, given that such a high percentage of caregivers indicate dissatisfaction with their medical team, as evidenced in previous research (Daily et al., 2016; Cheuk et al., 2004; Wei et al., 2016) that there is a general lack of communication about QOL issues (Marino et al 2012; Marino 2010). Results indicated over 70% of caregivers reported neurodevelopmental disability risks and screenings and mental health risks not being adequately addressed. Implications suggest a great need for cCHD providers and family integrated care to address the risks and prevalence of neurodevelopment and mental health issues that may arise/exist.

Implications

Research exploring issues related to family and children's well-being when children have a chronic illness such as cCHD is limited. This research contributes to this gap in research by providing a novel examination of how parents' knowledge and satisfaction with their child's medical team impact overall family and child well-being. Congenital heart disease affects more families than any other birth defect in the world. Rising morbidities associated with cCHD emphasize the importance of empirical data and information that may help the vast number of

cCHD families and children that are struggling to find answers and do not know how to obtain the resources to provide the knowledge they seek, or that shared knowledge is available.

Limitations

The current study had several limitations. The participant sample size was small in relation to the proposed sample size for this research. Initial recruitment expectations were based on an earlier study where posting a digital recruitment flyer resulted in participant responses from over 90 cCHD caregivers. However, participant eligibility in the present study were limited by the inclusion criteria that caregivers have a child with cCHD five years or older and has attended school or an education based program. This would limit the eligible participant pool significantly in comparison to the previous research study. Additionally, and perhaps more relevant to limited participant response was that recruitment occurred during the initial stay-at-home orders experienced in the United States in early 2020. With this experience, families were likely focused on family cCHD needs. The uncertainty of the time and shifting responsibilities of parents may have precluded eligible caregivers' participation.

When there were few new participants over a period of time, with multiple study recruitment attempts, the study concluded. Analyses with available data were conducted to determine whether the number of participant responses collected to that time would allow a valid analysis. Analyses addressing the small sample size indicated the current findings were not impacted, having a larger sample size may have brought more in-depth knowledge and statistically significant findings. A larger sample size would have increased the power of the analysis and future studies should increase sample size to increase power and would potentially validate current findings and find statistical findings in areas that were not found to be statistically significant in the current study.

Although the sample would support analyses, the sample was homogeneous in nature, race and sex. For example, only mothers that identified as White responded to survey and also met the criteria for participation in study. It is possible that by conducting data collection during the COVID-19 pandemic, the participant homogeneous sample may have been influenced by economic hardships, single parent homes, childcare factors, i.e. health disparities that often impact black and brown communities more significantly than their White counterparts. In addition, the pandemic may have potentially influenced scores and responses reported for scales assessing mental health, specifically PTSD and depression.

Future Research

Future research would benefit from examining QOL and caregiver knowledge and nonclinical factors. In addition, obtaining participants within a doctor or hospital setting will increase sample size and diversity.

Caregiver knowledge of risks associated with cCHD as a possible influencing factor is highly important and a significant valued outcome variable. The current research study helps lay the foundation for future cCHD family and physician research and interventions, to focus on increasing healthy family functioning and building resilience within the cCHD community. The results of the study indicate and support the importance of continued research related to medical team—family interactions indicating continuing the examination in future research studies and within healthcare provider settings. The implications suggest caregiver knowledge of cCHD risks (neurodevelopmental delays, mental health, and provider quality care) have a statistically significant positive relationship with caregivers' satisfaction with their healthcare provider (treatment and care approach). Results indicate the provider treatment care approach significantly relates to caregiver knowledge, indicating potential associations with other areas

that pose risks may also be associated to caregiver knowledge.

The indication of caregiver knowledge of cCHD risks influencing the type of provider care and approach is significant, bringing many potential new interventions and a deeper understanding on how to best serve the needs of children with cCHD and their family. The provider care approach is an area that has received increased attention by advocates and cCHD health organizations, yet needs additional exploration in cCHD family research. The recent guidelines for providers to use a psychosocial integrated family center care approach is groundbreaking. The communication and knowledge gap that exists between providers and caregivers can greatly close by this type of approach, resulting in higher levels of caregiver cCHD knowledge, and access to resources and interventions for the cCHD family. Using a holistic integrated approach by the cCHD healthcare provider could open doors for pediatric collaborative family health, offering mental health, neurodevelopmental screenings, and other needed assessments to the child with cCHD and family all in one location. educate and provide knowledge to children with cCHD and families.

To best provide support to parents whose children have cCHD, it is important to move beyond the basic understanding that caregiver knowledge of potential risks to their child's QOL outcome—as well as the QOL outcome for the family as a whole. Furthermore, research indicates families that are knowledgeable of cCHD diagnosis, caring for their child, are able to re-adapt and unite together report gaining strength and healthier overall family functioning than before the diagnosis (Lubkin & Laresen, 2013; Uzark, et al., 2003) Reliable information for cCHD caregivers is scarce and resources often remain unknown. The American Heart Association conducted a study focused on parental knowledge and children with cCHD's future needs. Results indicated that over half of the cCHD caregivers were found to have limited or

incomplete knowledge regarding the child with cCHD's future needs and overall long-term care. More than 80% of those caring for a child with cCHD desire more education and knowledge regarding their child's illness, prognosis, and long-term quality care and only 13% cited informational resource sources other than their cardiac healthcare provider (Koch & Jones, 2018).

Understanding how parents' knowledge of the developmental trajectories and risks experienced by children with cCHD impacts families' and children's QOL is a first step to understanding the importance and value of integrated, systems-based care. As the findings could potentially emphasize the importance of integrated health care between cCHD providers and the family, specifically mental health. Is it possible that the majority of participants who scored low on the LKQ-CHD and lack knowledge, and by not having a large amount of depth of understanding of what risks are at risk is associated to higher scores on QOL? The LKQ-CHD revised form for the pediatric population as used in the current study should continue to be used in future research to assess knowledge of caregivers and further assess statistical validity and reliability. The scale could play an essential role for providers to use with families to help.

Research with a focus on how different approaches to care impact families and children's health can further inform the potential benefits of integrated health care and the role of caregivers' satisfaction with care providers. Future research should focus on the provider care approach in depth, assessing cCHD caregivers and physicians, and explore additional cCHD family needs associated with health disparities and inequalities. Work based on qualitative methodologies will provide more in-depth, rich data providing insight into the lives and experiences of children with cCHD and their families. By utilizing a collaborative, integrated healthcare approach inequalities and injustice become shared knowledge, caregiver knowledge

of risks associated with cCHD and available resources to healthcare increase, and healthcare providers can implement more effective interventions within cCHD families by use of holistic approach between caregivers and providers. This allows for researchers to not only address the *problem*, or risks, but also address intervention. This is significant due to current research within CCHD risks and inequality focusing only on the problem but not implementation of how to address problems and intervene.

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APPENDIX A

DESCRIPTIVE ANALYSIS QUESTIONS

Approximately 71% of caregivers reported that information regarding risks of neurodevelopmental delays, and thereby the need for neurodevelopmental screenings, are not adequately addressed in relation to implications for the child with cCHD or knowledge for the caregiver (none-minimal). Over 80% of caregivers reported mental health risks were not being adequately addressed for either in relation to the child with cCHD, or regarding the caregiver's knowledge of the risks (none-minimal).

	Frequency	Percent	Valid %	Cum %
How often has your child's health provider discussed mental health related issues?				
A lot	3	6.5	6.5	6.5
A moderate amount	6	13.0	13.0	19.6
A little	14	30.4	30.4	50.0
None at all	23	50.0	50.0	100.0
Total	46	100.0	100.0	
How has your child with cCHD's health provider discussed mental health related issues with you as potential issues for you?				
A Lot	2	4.3	4.3	4.3
A moderate amount	4	8.7	8.7	13.0
A little	10	21.7	21.7	34.8
Not at all	30	65.2	65.2	100.0
Total	46	100.0	100.0	
How often has your child with cCHD's health provider discussed neurodevelopmental disabilities with you as potential for your child with cCHD?				
A lot	3	6.5	6.5	6.5
A moderate amount	10	21.7	21.7	28.3
A little	15	32.6	32.6	60.9
Not at all	18	39.1	39.1	100.0
Total	46	100.0	100.0	
How often has the need for neurodevelopmental screenings been discussed with you for your child with cCHD by healthcare provider?				
A lot	3	6.5	6.5	6.5
A moderate amount	10	21.7	21.7	28.3
A little	17	37.0	37.0	65.2

	Frequency	Percent	Valid %	Cum %
Not at all	16	34.8	34.8	100.0
Total	46	100.0	100.0	

APPENDIX B

EXTENDED LITERATURE REVIEW

Theoretical Frameworks

Chronic illness in a child, such as cCHD, has been problematized within conceptual frameworks guided by family systems theory and ecological systems theory (Kazak& Nachman, 1991). The diagnosis of cCHD cannot be understood as an isolated factor or as impactful to the child alone. Family systems theory posits that the parts, or members, within the family system are interconnected, and true conceptualization requires viewing the family structure as a whole (McDaniel, Doherty, & Hepworth, 2013). Bronfenbrenner's ecological systems theory is vital within cCHD family research due to the interaction between the cCHD family.

Extended Literature Review

The microsystem (child with cCHD), mesosystem (family, school, cardiac medical provider), and exosystem (hospital, extended medical providers, AHA, healthcare policy), and macrosystem (attitudes and ideology of culture regarding cCHD and chronic illness) that remains constant, and can serve as protective/adaptive functioning within the cCHD family. In addition, the family system and functioning ability influences the environment, as environmental factors influence the cCHD family (McCusker & Casey, 2016).

A cCHD diagnosis has a direct impact on each family member, instantly changing the structure of the family. Barlow and Ellard (2004) refer to childhood chronic illness as a "family affair", and emphasizes the need to treat the whole family, specifically the family being part of the medical treatment team. The family roles, responsibilities, structure and beliefs typically change as a result of the illness. Research indicates families that are able to readapt and function/unite together report gaining strength and healthier overall family functioning than before the cCHD diagnosis (Lubkin & Laresen, 2013; Uzark, Jones, Burwinkle, & Varni, 2003).

Knowledge of the cCHD illness itself, potential risks, and interventions to address risks are vital. A vast amount of current research involving chronic illness in children are based on implementing family system interventions, that focus on providing knowledge, empowerment, and resources needed for adaptability and healthy family functioning (McDaniel et al., 2013).

A family with a child diagnosed with cCHD involves constant transactions across the four levels of Bronfenbrenner's ecological systems. Protective and risk factors can have a direct influence on capabilities and demands and all can occur at different levels within the ecological system (McDaniel et al., 2013). The cCHD family can be negatively impacted during transactions across the ecological systems. Families needing cCHD resources and knowledge can counteract negative transactions across the ecological systems by allowing the system to be an advantage, a resourceful life changing transaction, but the vast gap in cCHD research on risks associated with cCHD must be addressed (McCusker & Casey, 2016). Bronfenbrenner's ecological systems theory intertwines simultaneously with the family systems framework in family medical research and, together, form the conceptual framework for the proposed research study.

Chronic Illness in Children and Family Impact

Having a child with cCHD can be very stressful for parents, facing overwhelming emotions and extra physical, financial, and other practical challenges. Caregivers are at risk of psychological distress, anxiety, depression, somatization, hopelessness, and posttraumatic stress symptoms, which in turn may influence caregiver's responsiveness. A holistic approach to early biopsychosocial interventions should aim at improving coping, enhance parenting, and family functioning. During routine medical checkups, medical professionals should ask about parental

stress, family functioning, and psychosocial functioning of the child and, when needed, adequate biopsychosocial care should be provided (McDaniel et al., 2012).

"Well-Siblings".

Siblings of a child diagnosed with cCHD are often overlooked by researchers, caregivers, and the medical team. Research that has been conducted regarding the well sibling indicates that the diagnosis has a drastic and life changing impact on their life projector (Wei, Roscigno, Hanson, & Swanson, 2015). Often more responsibilities related to care and household responsibilities are required during the ill sibling's hospitalizations and at home aftercare recovery periods (Steltzer, 2016). Additional aspects related to a sibling cCHD diagnosis that impact the well sibling include fear of the unknown, uncertainty of potential death, wellness/survivor's guilt, feeling unimportant, behavioral/mental health issues, and anger as a result of having to frame the family foundation and activities around the current heath state of the child with cCHD (Steltzer, 2016; Wei et al., 2015). There is a need to emphasize the importance of developing programs that address the well – sibling during childhood, as well as throughout adulthood. It is essential for future research to focus on the impact that cCHD has on the well sibling to fully understand the interventions that need to be developed and implemented for healthy life development (Steltzer, 2016).

Absence of the Father Figure

There is a deficit within the literature pertaining to father's experiences of having a child diagnosed with congenital heart disease. The majority of research has excluded the experiences of father's caregiving role (Gower, Higgins, Doherty, & McCormack, 2017). Prior research indicates that often the mother of a child with chronic illness is the sole focus of studies that address the experience of caring for a child with a chronic illness. However, recent research

indicates that mother's experiences are much different than a father's experience. Fathers often feel they are not recognized by the medical team as part of the treatment team, or had a say in the child's care/treatment as much as would have been preferred (Gower et al., 2017). Fathers influence the family structure. In addition, father's role in caretaking has drastically increased over the last several decades, therefor it is essential to understand father's experiences of caring for a child with congenital heart disease (Gower et al., 2017; Wei et al., 2015).

Risks Associated with Families with a Child Diagnosed with cCHD

Caregiver Knowledge of Risks

Caregiver knowledge for the current study is defined as knowledge the caregiver has regarding family risks associated with having a child diagnosed with cCHD. Specifically, potential risks and risks of neurodevelopmental disabilities and mental health. Caregiver knowledge for the current study is defined as knowledge the caregiver has regarding risks associated with having a child diagnosed with cCHD family. Thus, directly impacting the cCHD family and child's QOL. The focus is commonly centered around cCHD medical related risks (future surgeries, activity limitations, current cardiac functioning), with minimal focus on potential future risks outside the cCHD diagnosis being discussed with and/or considered by the cCHD caregiver (Marino et al., 2012).

Caring for a child with cCHD is associated with a wide range of challenges for families and the child with cCHD. Parent knowledge and perception of the child with cCHD's illness are positively correlated with child with cCHD developmental outcomes, and family coping/adaption (Wang, Hay, Clarke, & Menahem, 2014; Garcia, Aggarwal, & Natarajan, 2016; McCusker & Casey, 2016). Understanding a child's illness enhances competency in the caregiver, lowers stress/anxiety levels, and directly impacts the overall mental and physical

wellbeing and development of the child with cCHD. Caregiver cCHD knowledge increases feeling of control, self-efficacy in caring for child with cCHD, and reduces thoughts of uncertainty. Uncertainty of how to care for the child with cCHD, what to expect/prognosis are significantly guided by feelings of uncertainty and anxiety are highly reported amongst cCHD caregivers (Koch & Jones, 2018).

Mental Health and Trauma

Mothers of children recently diagnosed with CHD often report feelings of hopelessness, depression, anxiety, and isolation. While mother's report feelings of post-traumatic stress disorder and ruminating worrisome thoughts as life-long symptomologies (Bruce et al., 2014). Children diagnosed with CHD, and their siblings have high incidences of anxiety, social exclusion, and exhibiting behavioral and emotion regulation issues, siblings also often report feelings guilt and resentment (Sabzevari, Nematollahi, Mirzaei, & Ravari, 2016; Wei et al., 2015).

Many caregivers experience a period of grieving when first learning of their children's diagnosis. Parents grieve the loss of the life that had been envisioned for the CHD child and grieving as they come to terms with the reality and uncertainty of their child's life (Sabzevari et al., 2016). The importance of supporting parents' through this process is evident in the literature. Caregiver poor mental health also is associated with caregiver burnout and is prevalent amongst those caring for a child with cCHD. Reports of burnout are highest for both single caregiver households and for caregivers with a partner that does not co-parent effectively, neglecting daily caregiving responsibilities (McDaniel et al., 2013).

A caregiver's lack of knowledge and awareness regarding their child's diagnosis, prognosis, risks, and effective interventions evokes feelings of anxiety and high chronic stress in

families, and has a direct association to mental health wellbeing. This restricts the ability for resiliency to develop, a vital component for adaptive family functioning (Jackson, Liang, Frydenberg, Higgins, & Murphy, 2016; Patterson, 2003).

Developmental Disabilities and Psychosocial Delays

Once children with cCHD began attending school, it became apparent that neurodevelopmental delays were present in more than half of the children with cCHD in elementary school (BCHCB, 2017; McCusker & Casey, 2016). Psychosocial delays were also identified, and are considered a comorbidity of neurodevelopmental disabilities. Neurodevelopmental delays in the child with cCHD range from manageable to significantly impaired. Providers treating cCHD were not prepared for the neurodevelopmental and psychosocial risks that children are experiencing, as it is unexpected, and the cause not clearly understood (Shillingford et al., 2008).

The American Heart Association (AMA) released the first scientific statement clearly identifying the high risk and prevalence of neurodevelopmental and psychosocial impairments within the cCHD population. The formal statement provided cardiac providers with a step by step guideline for surveillance and treatment management to be implemented with all cCHD patients. Guidelines reported by the AMA included a recommendation for cardiac providers to discuss a “crisis” (Marino, Cassedy, Drotar, & Wray, 2016).). One of the main objectives for the formal statement release was to signify how early intervention was vital in the decrease of long-term developmental delays and increase in long-term QOL (LTQOL), (Marino et al., 2012). The negative impact and implications of risks being unaddressed/undiagnosed and delayed in diagnosis are associated with significantly lower academic and socio-emotional functioning, thus impacting the overall QOL for the child with cCHD and family.

Academic and Socio-Emotional Functioning

Significantly high prevalence in both academic and socio-emotional ability/functioning reports in children with cCHD highlights the urgent need to assess and understand protective, proactive environmental influences, such as the active role of the caregiver and caregiver. knowledge Marino (2012) highlights the vital role and impact of parent wellbeing on a child's development (health, neurodevelopment, and socio-emotional), and the significance of modeling healthy coping strategies. Caregivers and the family environment are the most important predictor for children with cCHD's QOL and set the trajectory for the child with cCHD's developmental outcomes, including academic success and socio-emotional competence.

Caregiver's awareness of psychosocial impairments were associated with success in academia peer acceptance, increases early intervention implementation and higher quality parent-child relationship (Marino, Cassedy, Drotar, & Wray, 2016). A recent study found that over half of cCHD caregivers rate themselves at adequately informed regarding their child with cCHD's long-term care, yet less than half have objectively sufficient knowledge. Although caregiver knowledge relating to cCHD risks/risks and developmental outcomes are limited, mother's involvement has been identified as an influential factor in identification and early diagnoses of developmental deficiencies, early implementation of interventional methods, and decreases in additional developmental delays (Brosig et al., 2017).

cCHD Caregiver-Provider Relationship: Interactions across Ecological Systems

The provider-caregiver relationship/treatment model is defined by the quality, communication, interaction, and treatment of care approach associated with the cCHD illness that exists between the child with cCHD's cardiac medical provider and the child with cCHD's caregiver (O'Conner et al., 2016). Existing research on cCHD as it relates to the field of family

science and an integrated holistic provider approach presents a major gap within the available literature, limiting the amount of resourceful information available to cCHD caregivers (Daily et al., 2016). The American Heart Association conducted a study focused on parental knowledge and children with cCHD's future needs. Results indicated that over half of the cCHD caregivers were found to have limited or incomplete knowledge regarding the child with cCHD's future needs, risks and overall long-term care. More than 80% of those caring for a child with cCHD desired more education and knowledge regarding their child's illness, prognosis, and long-term quality care and only 13% cited informational resource sources other than the cardiac medical provider treating their child with cCHD (Miller, 2018).

Physicians agree that caregivers are insufficiently involved in the cCHD care and treatment, and cite a lack of comprehension as the cause, yet cardiac physicians report to conveying information to caregivers mainly based on their own judgment of what is important/comprehensible to the caregiver (Lubkin & Larsen, 2013). This is a major barrier to the lack of parental knowledge within the cCHD population as information provided by the physician may not always align with what caregivers think are important (Etnel et al., 2017).

Shared Knowledge, A Family-Center Psychosocial Care Approach

It is often reported that caregivers report a lack of communication and feeling understood by the medical team (Woolf-King et al., 2018). Feelings of confusion and being misunderstood by the medical team is an issue that can arise between the medical team and the caregiver (Sabzevari et al., 2016; Wei et al., 2015). The challenges and risks experienced by families of a child diagnosed with cCHD contribute to tension, stress, dysfunction, and mental health issues and trauma. The overall implications for the inconsistency in cardiac quality care for health disparities and risks associated with cCHD and the family are multifaceted. The issues are

grounded in education, training and general awareness of the comorbidities that exists between cCHD and healthy family functioning (Cassidy, 2017). Healthcare provider's treatment recommendations for screening/monitoring potential risks and risks for cCHD vary considerably. It is essential that these challenges and risks are more clearly understood for appropriate intervention methods to be developed and implemented (Wei et al., 2015).

The family-centered psychosocial care approach aims to strengthen the resilience of cCHD families by adopting a holistic approach for the whole family, body and mind perspective (Utens, Callus, Levert, Groote, & Casey, 2018). Long term cCHD developmental outcomes and QOL are significantly higher for the child with cCHD and caregiver with a cardiac medical provider that implements a provider-caregiver shared medical treatment team approach. Developing new interventions to access and provide resources for the cCHD family associated risks is vital for QOL and overall family functioning.

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