AN URBAN BIOETHICS APPROACH TO UNDERSTANDING DISPARITIES IN NEURODEVELOPMENTAL OUTCOMES FOR CHILDREN WITH CONGENITAL HEART DISEASE

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ABSTRACT

Congenital heart disease (CHD) is the most common birth defect and often results in neurodevelopmental impairments and psychological problems which impede educational and occupational attainment and decrease overall quality of life into adulthood. While morbidity and mortality outcomes have improved over the last several decades, non-Hispanic black and Hispanic children continue to experience a disproportionate burden of CHD. An urban bioethics approach to disparities in cardiac neurodevelopmental outcomes necessitates an examination of the context, setting, and structures in which CHD care is delivered. This thesis proposes a model through which *access to* and *quality of* cardiac care impact disparities in neurodevelopmental outcomes. The thesis describes an initial evaluation of the proposed model conducted through retrospective record review. Though research funding and hospital resources have historically flowed toward optimizing surgical and other clinical care techniques, results indicate that factors such as poverty and other social determinants of health have a greater impact on many CHD outcomes. An urban bioethics framework asks us to additionally consider the ways in which cardiac care teams act as barriers to high quality care. Findings are discussed in terms of next steps and a proposed qualitative study to further evaluate results.

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CHAPTER 1: INTRODUCTION

Congenital heart disease (CHD) is the most common birth defect, affecting nearly 1% of children worldwide (van der Linde et al., 2011), and the most common cause of birth defect related mortality (Gilboa et al., 2010). Approximately 25% (Reller et al., 2008) of children with CHD have a complex form of the disease, requiring surgical intervention within the first year (and often first month) of life. Early surgery and extended hospitalization results in exposure to noxious stressors in the hospital (e.g., needle sticks, bright lights, loud noises), reduced opportunities for normative developmental experiences, and prolonged separation from parents during a highly vulnerable time when bonding and attachment typically occur (Carbajal et al., 2008; Lisanti et al., 2019). These early experiences, along with the cerebrovascular physiology and neurological sequelae of CHD, place children with CHD at high risk for neurodevelopmental impairments (i.e., developmental delays, autism spectrum disorders, and attention-deficit/hyperactivity disorder) and psychological problems (i.e., depression and anxiety; Verrall et al., 2019; Wernovsky & Licht, 2016) which impede educational and occupational attainment and decrease overall quality of life into adulthood.

While morbidity and mortality outcomes have improved over the last several decades, non-Hispanic black and Hispanic children continue to experience a disproportionate burden of CHD. Mortality resulting from CHD has declined almost 40% in the last 20 years, however, the rate of change in mortality outcomes has been significantly greater for non-Hispanic white as compared to non-Hispanic Black and Hispanic individuals (Lopez et al., 2020).

An Urban Bioethics Approach to Disparities in CHD Outcomes

An urban bioethics approach may help us understand what drives and maintains these disparities. Urban bioethics is a branch of bioethics which considers the context in which bioethical decision making occurs (Blustein & Fleischman, 2004). It is particularly relevant when medical decision making takes place in settings that are dense, disparate, and diverse. Infants with critical CHDs almost always receive surgical and post-surgical care at children's hospital in urban settings. Thus, their formative, earliest experiences in life and in medical care are centered in density, disparity, and diversity.

Urban bioethics provides us with three tools we can apply to ethical analysis: *Agency* refers to the ability, capacity, and true opportunity to make choices from a complete range of options, in contrast to the traditional bioethics concept of autonomy, which refers to the ethical ideal of freely made choice; *Solidarity* refers to the principle of binding stakeholders together and implies that both stakeholders work toward a common goal, rather than a hierarchical framework of physician determined beneficence/non-maleficence; *Social justice* asks us to consider the structures in place that impact inequities in the distribution of resources and care, rather than the traditional notion of justice as acontextual fairness and equality.

With regard to CHD outcome disparities, these three tools necessitate that we examine the context, setting, and structures in which CHD care is delivered. Incidence of CHD does not vary by race or ethnicity (Egbe et al., 2014), indicating that disparities in care following diagnosis account for these mortality trends. The overall improvement in CHD outcomes is due to tremendous innovations in surgical techniques and clinical care

during the neonatal period and infancy through international quality improvement collaboratives and due to the advocacy efforts of parents and families (Hickey et al., 2017; Jacobs, 2015). Thus, the persistence of health disparities for children with CHD based on race and ethnicity points toward inequity in the implementation of surgical innovations and improved care techniques.

Social determinants of health contribute to the relation between patient sociodemographic characteristics and clinical outcomes for children and adults with CHD (Davey et al., 2021). Social determinants of health (SDH) refers to the conditions and structures that surround people through development and daily life that impact health and quality of life outcomes (US Department of Health and Human Services, 2010). These determinants are typically grouped into economic stability, education access and quality, health care access and quality, neighborhood and built environment, and social and community context. SDH impact outcomes beginning at diagnosis. Families from lower income households are much less likely to receive a CHD diagnosis prenatally (Hill et al., 2015; Peiris et al., 2009; Purkey et al., 2019). Because they learn about the diagnosis during the postpartum period, these families are less likely to be prepared financially or emotionally. Families who already have fewer resources bear a larger economic burden of CHD. With less time to plan for and learn about CHD, they are less likely to seek out and receive developmental services after their child is born. After diagnosis, poverty, insurance barriers, limited access to transportation, and immigration status are all associated with infant mortality (Almli et al., 2017; Fixler et al., 2012; Kucik, Cassell, et al., 2014; Kucik, Nembhard, et al., 2014; Pace et al., 2018; Peyvandi et al., 2018; Siffel et al., 2015). Poverty is consistently associated with post-surgical outcomes as well,

including transplant-free survival rates (Tashiro et al., 2014), unplanned readmissions (Lushaj et al., 2020), and length of hospital admissions (Peterson et al., 2017).

Disparities in neurodevelopmental outcomes related to the social context of care have been identified as well. Children with CHD who live in neighborhoods with lower average household income tend to experience motor and cognitive delays, more difficulties with adaptive skills, and worse social, emotional, and behavioral functioning, with gaps in development between the highest and lowest neighborhood income groups widening over time (Bucholz et al., 2020, 2021). An extensive body of literature has identified perioperative factors that impact cardiac neurodevelopmental outcomes, including length of cardiopulmonary bypass, use of specific surgical techniques, and exposure to deep hypothermic circulatory arrest (Ligsay & Goldberg, 2021; Marino et al., 2012). However, research also indicates that patient factors, including sociodemographic characteristics, are more important in predicting developmental outcomes for children with CHD compared to perioperative factors (Goldberg et al., 2019). Improvement to these preoperative factors, though crucial to ensuring the survival of patients, is less likely to impact complex, long-term outcomes such as problem-solving abilities or social skills. Disparities in cardiac neurodevelopmental outcomes have likely flourished due to the narrow focus on perioperative factors with little consideration to the social context in which cardiac care is provided.

These results speak to the profound impact that lack of access to high quality care has on mortality and morbidity outcomes. In the long term, poverty, transportation barriers, and immigration status continue to impact the likelihood of accessing cardiac

care on the recommended schedule (Lu et al., 2017; Mackie et al., 2012). In terms of neurodevelopmental outcomes specifically, poverty and transportation barriers are associated with delayed cognitive development, lower grade-level literacy and math proficiency, and greater difficulties with adaptive behaviors (Bean Jaworski et al., 2017; Majnemer et al., 2008; Mulkey et al., 2016). One study has found that regardless of CHD severity, lower income predicts lower family quality of life (Lee et al., 2020). Travel distance to a specialized cardiac care center also appears to impact the chances of a child with CHD achieving literacy at grade level (Mulkey et al., 2016).

Provider Factors and Quality of CHD Care

While CHD research has just begun to identify factors associated with health disparities, primarily through quantitative studies, we can look to literature on health disparities among premature infants treated in neonatal intensive care units (NICUs) to suggest additional drivers of inequity. Infants cared for in NICUs tend to share similarities with infants with CHD cared for in cardiac intensive care units (CICUs). Though a more heterogeneous group, infants cared for in NICUs are also at higher risk for neurodevelopmental delays and have benefitted from neuroprotective interventions aimed at promoting family-centered care and better aligning the NICU setting with that of a typical newborn environment (Lisanti et al., 2019; Phillips, 2015). Thus, NICU literature may point toward next steps in research and intervention to address disparities in health outcomes for children with CHD.

NICU literature suggests that while SDH and access to care impact outcomes, so too does the quality of care provided (Beck et al., 2020). “Disparities in quality of care" is

defined as the health disparities that arise along racial and ethnic boundaries that are not due to access-related or other clinical factors (Profit et al., 2017). Infants of color are more likely to receive care at NICUs with access to fewer resources, and to subsequently receive lower quality of care, reflecting structural inequalities (Howell, 2008). Infants of color are also more likely to receive lower quality of care within individual centers, however, reflecting organizational and clinical inequalities due to racism and discrimination. One qualitative study found that mothers of infants admitted to the NICU at a safety net hospital experienced barriers to participating in infant care during hospitalization, including inadequate maternity leave and difficulties with housing, transportation to the hospital, and care of siblings (Lewis et al., 2019). These factors prevented the mothers interviewed from spending time at the bedside during their infant's NICU admission and from providing hands on care including skin-to-skin contact and breastfeeding—interventions known to improve outcomes for premature infants. Within the same care center, infants of parents who report lower socioeconomic status are then less likely to receive the highest quality care to ensure optimal health and neurodevelopmental outcomes.

Beyond patient and family resources, provider factors are a major contributor to inequitable care in NICUs. One qualitative study found that, for mothers who reported lower socioeconomic status, consistency of information received from their infant's medical team threatened their ability to trust and willingness to engage with providers in their infant's care (Enlow et al., 2017). Sigurdson and colleagues (2018) identified two primary provider factors that drive suboptimal outcomes in the NICU: neglectful care (i.e., providers ignore or avoid certain families, identify families as difficult or

unpleasant, and neglect to provide supports needed to optimize infant outcomes) and judgmental care (i.e., providers hold judgmental or discriminatory attitudes toward families based on family cultural practices and beliefs, race, class, or immigration status). Families are acutely aware of these attitudes and behaviors. Parents of color and of low socioeconomic status report major disruptions in power sharing and trust during NICU admissions due to judgmental, discriminatory, and racist actions and attitudes from care team members (Sigurdson et al., 2020). While similar research conducted in the CICU with CHD patients and families does not yet exist, my observations as a provider are consistent.

CHAPTER 2: DISPARITIES IN NEURODEVELOPMENTAL OUTCOMES

A review of the CHD literature reveals gaps in both the content and production of research concerning disparities in long-term outcomes for children with CHD. In their framework for addressing disparities in healthcare settings, Kilbourne and colleagues (2006) recommend that, after disparities have been identified, research agendas should prioritize understanding the mechanisms that drive disparities so that interventions to reduce these disparities can be developed. As summarized above, the disparities have been identified. Few studies have identified the mediators that link family characteristics to disparate outcomes, limiting progress toward effective interventions. In terms of production, research in this area has primarily centered the experiences and expertise of the physician or other cardiac care team providers. Limited research has incorporated family and patient perspectives (Sood et al., 2021). Among CHD outcomes research that has included family perspectives, engagement of diverse voices, namely, families who identify has Black or Hispanic, has not been a primary goal. Research that ignores the perspective of the population impacted by the research is unlikely to be effective, particularly in the case of interventions research where buy-in is essential.

To map a road forward toward the development of interventions that reduce health disparities in CHD care, a model is proposed (Figure 1). The model includes possible mechanisms through which some patients with CHD are at greater risk for neurodevelopmental delays, as well as avenues to engage the lived experiences of patients and families most impacted by the identified disparities. The model begins with three major areas identified as modifiable factors that impact neurodevelopmental

outcomes in CHD (Ryan et al., 2019): parent engagement in the CICU environment, parental mental health, and access to long-term follow up care.

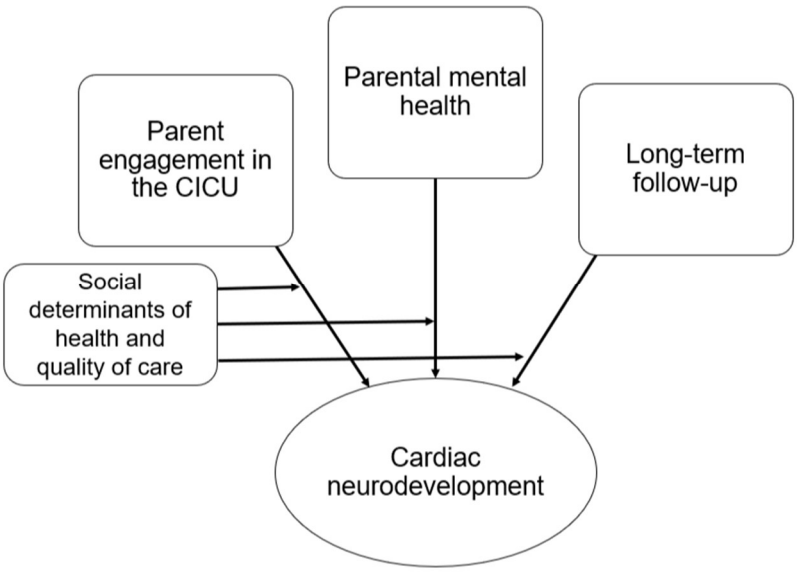

Figure 1. The Intersection of Access to High Quality Care with Predictors of Neurodevelopmental Outcomes for Children with CHD

Parent Engagement in the CICU

Infants admitted to the CICU following cardiac surgery are hemodynamically unstable and must be separated from parents during a critical period for bonding and attachment (Lisanti et al., 2017). Parents are often limited in their ability to perform normative parenting tasks such as feeding, diapering, holding, and soothing their infant. The CICU environment is at odds with the typical home environment, and infants are subjected to noxious and painful stressors including diagnostic testing and critical care interventions that disrupt their ability to rest, sleep, and heal (Daniels & Harrison, 2016). Evidence has accumulated that the combined burden of surgery during the neonatal

period, exposure to pharmacological therapies, sleep disruption, and separation from caregivers has long term effects.

Though life-saving, the CICU environment, and the interventions infants receive when admitted, contribute to long term alterations in neurodevelopment. The longer infants are admitted to the CICU after cardiac surgery, the greater impact on cognitive functioning in childhood, likely due to the greater cumulative exposure to CICU interventions (Wernovsky & Licht, 2016). While close contact with their caregivers supports nervous system maturation and development of self-regulatory skills, infants with CHD who are typically separated from caregivers in the CICU environment may fail to overcome autonomic dysregulation associated with the pathophysiology of CHD, impacting neurodevelopmental outcomes through adolescence (Harrison & Brown, 2017; Mulkey & du Plessis, 2019). Parent engagement buffers this effect both directly (through skin-to-skin care and similar practices which reduce infant stress and dysregulation) and indirectly by supporting parents in learning to provide responsive care for their child's unique medical needs and to be advocates for this needs during and after hospitalization (Lisanti et al., 2019).

Parental Mental Health

Separation from caregivers in the CICU disrupts opportunities for non-pharmacologic stress reduction for infants, and contributes to increased stress for parents as well. Anxiety, depression, and other post-traumatic stress symptoms are common among parents of children with CHD (Woolf-King et al., 2017). Parents cite numerous additional stressors that contribute to these mental health symptoms, including learning of

their child's diagnosis, separation from their child after birth for surgical intervention, seeing their child intubated and sedated after surgery, and watching their child or other children in the CICU be resuscitated (Sood et al., 2018). Parents who experience anxiety, depression, and post traumatic stress during the neonatal period are likely to continue experiencing these symptoms long term. While psychosocial treatment options for parents during and after their child's hospital admission are increasing, there is still a tremendous need for recognition of the importance of addressing parent mental health as a core component of children's cardiac care and a need for providers adequately trained in evidence based treatments.

Parent mental health predicts neurodevelopment and behavioral health outcomes for children with CHD above and beyond clinical or surgical factors (McCusker et al., 2007). This effect begins in utero, with recent research demonstrating that maternal stress and anxiety during pregnancy predict reduced cerebellar and hippocampal volumes in infants with CHD after birth (Wu et al., 2020). During initial surgical hospitalization, stress and anxiety may make it difficult for parents to remain at the bedside with their child and engage in care (Sood et al., 2018). Many aspects of the CICU serve as trauma triggers for parents, who may avoid the setting as a maladaptive mechanism for coping with distress. Long term, parents who experience their own difficulties with behavioral health are less likely to have the emotional resources to responsively address their child's behavioral needs. Research indicates that parents who have endured the early trauma of witnessing their baby undergo major surgery may be less able to engage in the scaffolding behaviors that meet their child where they are and support them in developing new skills, contributing to developmental delays (Laing et al., 2010).

Long-Term Follow-Up

Long term support for medical and developmental needs comprises the third predictor of morbidity and mortality outcomes. The developmental differences children with CHD may experience can have large impact on their educational achievement and occupational attainment. School systems do not always have the means, capacity, or specialized knowledge to accurately assess the neurodevelopmental needs of a child with CHD and therefore may not be able to adequately tailor the learning environment to ensure children with CHD achieve to the best of their ability and experience success. Regular screening for developmental delays and behavioral health needs so that intervention can begin as early as possible has been the standard of care for children with CHD for the last decade (Marino et al., 2012). Regular medical follow up for surveillance and preventive medical care is also essential for reducing mortality and morbidity outcomes (Wren & O'Sullivan, 2013).

Access to and Quality of Care

Access to cardiac care and the *quality* of care once accessed are two factors which are proposed to impact each of the three domains described above. The SDH experienced by a family act as barriers to access, while provider factors (e.g., willingness and ability to engage in family centered care, provision of neglectful or judgmental care) act as barriers to high quality care. As described above, SDH significantly impact parents' ability to stay at the bedside with their child in the CICU and provide hands on care, to receive mental health care, and to routinely engage in long-term follow-up. Based on my own observations of cardiac care and a review of the NICU literature described above, it is hypothesized that parents whose infants receive low quality care in the CICU are less

able to spend time in the CICU and therefore have fewer opportunities to participate in medical decision making for their child. Parents who feel judged by providers or are not invited to participate in their child's care may over time be less likely to be present in the CICU due to anxiety, frustration, or general stress. They are then less knowledgeable about their child's care and less comfortable providing the care after discharge. This may further increase feelings of anxiety, depression, and stress through alterations to their expected parental role (Lisanti, 2018). Parents who are disengaged during their child's CICU stay may be less likely to receive psychosocial care from their child's cardiac team (i.e., informal check-ins about parents' mental health and empathic support) and be less likely to receive referrals to behavioral healthcare providers when needed. The slow erosion of trust between parents and their child's cardiac care team may then make parents less likely to schedule and attend recommended long-term follow-up care appointments.

An Initial Evaluation of Model Factors

With this model in mind, the following study was initiated to begin an examination of psychosocial factors that may contribute to disruptions in cardiac care for children with CHD. The purpose of this study was to examine both clinical and psychosocial factors to determine their relative contribution to disruptions to medical and neurodevelopmental care in the first two years of life. While the model presented here highlights the impact of psychosocial factors, including clinical factors in analysis will be important to situate the findings back into CHD literature, which has thus far invested in interventions targeting clinical care techniques, rather than psychosocial family centered care. The outcomes of interest selected for this initial examination of the proposed model

include variables related to inpatient cardiac care known to impact morbidity and mortality outcomes (i.e., length of stay, unplanned readmissions, and total number of readmission days) as well as variables related to outpatient care (i.e., adherence to recommended outpatient neurodevelopmental and cardiology appointments).

CHAPTER 3: METHODS

Participants and Procedures

Retrospective chart reviews were conducted for patients admitted to the Nemours Cardiac Center whose parent or caregiver completed the PAT between April 2016 and December 2019 as part of routine clinical care (n = 184). Parents/caregivers completed the PAT on a laptop computer brought to their rooms by bedside nurses. A best practice alert to administer the PAT was triggered in the electronic medical record for all patients under the age of 12 months admitted to either the CICU or the stepdown unit. Once completed, the psychologist, psychology fellow, and social worker for the cardiac center received an alert to review PAT responses. The PAT was only available in English for this study, so non-English speaking families were excluded. Patients were excluded from analyses if they did not have a primary diagnosis of CHD (e.g., supraventricular tachycardia) or were determined to have a social background that would confound analyses (e.g., multiple primary caretakers in the first two years of life) for a total sample size of 100 participants. This retrospective study was reviewed and approved by the Nemours Institutional Review Board with waiver of the requirement for obtaining informed consent/parental permission.

Variables and Measures

Family factors and patient clinical characteristics that may serve as predictor variables were extracted from the electronic medical record. These included race/ethnicity, living arrangement (e.g., lives with both parents, lives primarily with mother), maternal age and education, neighborhood poverty level using patient zip code (percentage of families living below the federal poverty level), distance from family

home to Nemours Cardiac Center and to their primary outpatient clinic in which they receive cardiology care, primary insurance, primary cardiac diagnosis and STAT score, diagnosis timing, gestational age, and presence of a genetic anomaly.

Outcome variables were also extracted from the electronic medical record for the first two years of life and included length of each admission, whether admissions were planned or unplanned, total days spent in the CICU and in the step-down unit for each admission, number of surgeries and catheterizations during each admission, whether families attended a neurodevelopmental assessment appointment in the first two years and adherence to recommended cardiology outpatient appointments in the first two years. Unplanned readmissions were defined as admissions to the cardiac center for any reason other than a scheduled procedure (e.g., illness). In-hospital monitoring after cardiac catheterization was not included as a readmission unless the hospitalization lasted longer than 24 hours. Admissions and surgeries clearly related to other medical conditions were also not included. Adherence to outpatient appointments was calculated by extracting the date of recommended follow up from after visit summaries and discharge instructions (e.g., 4 weeks after hospital discharge) and the date the family actually attended their follow up appointment. The difference between these dates was coded as the time of delay to follow up. These delays were then summed across the second year of life (i.e., from 12 to 24 months of age) for a final metric of overall adherence. If families attended a cardiology appointment earlier than recommended, this was coded as zero days delay.

The PAT is a brief screening tool designed to assess psychosocial risk factors for families of children diagnosed with serious childhood illness (Kazak et al., 2015). The

PAT, as originally developed, yields seven subscale scores: Family Structure and Resources, Family Problems, Social Support, Stress Reactions, Family Beliefs, Child Problems, and Sibling Problems. The PAT also yields a total score, which is stratified into three tiers of psychosocial risk: Universal, Targeted, or Clinical. The current study employed a version of the PAT developed specifically for hospitalized neonates and infants (e.g., patients in the NICU, CICU, or cardiac stepdown unit) which omits the Child Problems subscale given patient age. This version also includes items which are clinically relevant to the patient population but not included in any subscale or total score (e.g., Was the baby's hospitalization unexpected?).

Statistical Analyses

All data analyses were performed with SPSS Statistics, Version 27. Descriptive statistics (frequencies, means) were used to characterize the sample. Simple linear regressions were conducted between each predictor and outcome variable to establish initial estimates for baseline models. Variables with $p < .20$ were considered candidate predictors and entered into stepwise linear regressions to develop final multivariable models for each outcome of interest.

CHAPTER 4: RESULTS

Participants

Patients in this study were diverse with regard to race, ethnicity, and socioeconomic status (see Table 1). The majority of patients had public insurance (55.0%) and were living or planning to live with both biological parents following hospital discharge (83.0%). Mothers of patients in this study were 31.3 years on average (*SD* = 6.0) and were diverse with respect to education level. The percentage of the population living below the federal poverty line averaged across all zip codes for families in this study was 12.7% (*SD* = 7.5), which is comparable to the national poverty rate (https://www.census.gov/library/publications/2021/demo/p60-273.html). In terms of clinical characteristics, nearly a quarter of patients in this study had single ventricle anatomy (22.0%) and over half received a prenatal diagnosis of CHD (55.0%). Almost half had a documented genetic anomaly, including microdeletions (47.0%). The most common CHD diagnosis was ventricular septal defect (21.0%) followed by complete atrioventricular canal defect (14.0%) and Tetralogy of Fallot (10.0%). Most families' PAT Total scores fell within the Universal range (72.0%), with a quarter falling within in the Targeted range (25.0%) and 3.0% in the Clinical range, comparable to the initial validation study with this version of the PAT in the NICU.

Table 1. Sample Characteristics

	M (SD) or %
Family factors	
Poverty*	12.7 (7.5)
Race	
White	64.0%
Black	25.0%
Asian	2.0%
Other	9.0%
Ethnicity	
Non-Hispanic	80.0%
Hispanic	20.0%
Living arrangement	
Lives with both parents	83.0%
Lives with mother only	12.0%
Other	5.0%
Maternal age	31.3 (6.0)
Maternal education	
Some high school	8.0%
High school or GED	18.0%
Some college	26.0%
College or trade school	37.0%
Some graduate school	2.0%
Graduate school	7.0%
Distance to cardiac center	70.0 (131.7)
Distance to primary clinic	39.4 (56.5)
Primary insurance	
Public	55.0%
Private	40.0%
Tricare	2.0%
Self-pay	3.0%
Patient characteristics	
STAT score	1.2 (1.0)
Single ventricle anatomy	22.0%
Diagnosed prenatally	55.0%
Prematurity	22.0%
Gestational age	37.5 (2.7)
Presence of a genetic anomaly	47.0%
PAT scores	
Family structure	0.2 (0.2)
Support	0.03 (0.1)
Adult problems	0.1 (0.1)
Sibling problems	0.1 (0.2)
Stress reactions	0.08 (0.1)
Care beliefs	0.09 (0.09)
Total	0.6 (0.5)

*"Poverty" indicates percentage of the population below the poverty line

Analysis

Length of First Surgical Admission

The average length of first surgical admission was 52.4 days (*SD* = 75.9) including time spent in the CICU and cardiac step-down unit. Univariable analyses indicated that STAT score, single ventricle anatomy, prematurity, and gestational age were significant predictors of surgical admission length ($p < .05$). Distance to cardiac center ($p = .06$) and the PAT Adult Problems subscale ($p = .14$) were also included in multivariable analysis. STAT score was the only predictor included the final model, $R^2 = .082$, $p = .008$ (see Table 2 for all multivariable regression results). Unsurprisingly, the length of first surgical admission depended most strongly on the medical complexity of the child's CHD. While psychosocial factors may impact length of stay during this first surgery to some degree, medical factors are much more predictive.

Total Unplanned Readmissions

The average number of unplanned readmissions following first surgical admission was 1.1 (range: 0-9, *SD* = 1.9). Univariable analyses indicated that poverty, single ventricle anatomy, diagnosis timing, and the PAT Family Structure subscale were significant predictors of number of unplanned readmissions ($p < .05$). The PAT Care Beliefs subscale ($p = .15$) and PAT Total score ($p = .11$) were also included in multivariable analysis. The PAT Family Structure subscale and diagnosis timing were included the final model, $R^2 = .099$, $p = .003$.

Unplanned readmissions typically occur for this population when the child contracts a normal childhood illness (e.g., cold, flu) but due to their medical complexity

may have more difficulty breathing or become dehydrated more easily than their heart healthy peers. Thus, it is expected that children with CHD in this population on average had one unplanned readmission after their first surgical hospitalization. Unlike length of first hospital stay, psychosocial factors played a much larger role in determining which children would experience unplanned readmissions beyond the average. The PAT Family Structure subscale specifically refers to SDH including insurance status and access to reliable transportation to get to the hospital or outpatient appointments. That diagnosis timing emerged as a significant predictor likely reflects this variable as a proxy for family resources. As discussed in the introduction, families who experience barriers to reliably accessing high quality obstetric care are more likely to receive their child's CHD diagnosis postnatally. Thus, families who receive a postnatal diagnosis are likely also families who endorse higher PAT Family Structure subscale scores as well.

Total Readmission Days After First Surgery

The average number of readmission days following first surgical admission was 16.6 (SD = 38.9). Univariable analyses indicated that poverty, race, STAT score, single ventricle anatomy, and the PAT Family Structure subscale were significant predictors of total readmissions days ($p < .05$). Maternal education ($p = .08$), insurance type ($p = .19$), the PAT Adult Problems subscale ($p = .15$), and PAT Total score ($p = .05$) were also included in multivariable analysis. Poverty and single ventricle anatomy were included the final model, $R^2 = .22$, $p < .001$.

Poverty explained 21.9% of variance in the final model. Unlike the two prior outcomes examined, both medical and psychosocial factors predicted total readmission

days. However, children with single ventricle anatomy receive three planned surgical interventions. This was difficult to indicate in the database, and children with single ventricle diagnoses who would necessarily have more readmission days due to these additional planned surgeries were likely included in the count. It is surprising and unclear why neighborhood poverty level, rather than PAT Family Structure subscale score, was the unique predictor of readmission days. Children often spend more time in the hospital if they need to be discharged home with a feeding tube or other equipment as this requires parents to spend 24 hours on the unit receiving teaching and demonstrating that they can manage the child's medical care at home after discharge. For families who work long hours or are unable to take time away from work in the context of the financial strain of CHD, their children may remain in the hospital for additional days until they are able to receive discharge teaching. Poverty as a predictor may also reflect environmental factors, such as exposure to pollutants within a neighborhood, which can impact health outcomes and contribute to extended lengths of stay.

Attendance at Neurodevelopmental Follow-up

Univariable analyses indicated that the PAT Family Structure subscale, maternal education, distance to cardiac center, number of adults in the home, primary cardiac diagnosis, prematurity, and total CICU days were candidate predictors of whether a family attended a neurodevelopmental assessment. The PAT Family Structure subscale, total CICU days, and diagnosis timing were included the final model, $R^2 = .174$, $p = .011$. Children who spent more time in the CICU during their first surgical admission likely were not referred to their first neurodevelopmental follow up appointment as they remained admitted. This may reflect an area for improvement in our cardiac center's

referral process, as these children may not have been scheduled for subsequent evaluations after missing their first appointment. The PAT Family Structure subscale and diagnosis timing as predictors reflects the same factors that impact number of unplanned readmissions, namely, SDH.

Adherence to Recommended Cardiology Follow-up

Univariable analyses indicated that the PAT Family Structure subscale, the PAT Total score, STAT category, age at first surgery, and total cardiopulmonary bypass time were candidate predictors of adherence to recommended cardiology follow-up during the second year of life. The PAT Total score was the only variable included the final model, $R^2 = .084$, $p = .03$. It is unclear why the PAT Total score emerged as a predictor of adherence to cardiology follow-up but individual subscale scores did not. However, results indicate that psychosocial factors are the strongest predictor of whether a family is able to return for follow up on the schedule their child's provider recommends.

While SDH likely play a role here, as discussed above (i.e., families with access to fewer resources experience more barriers to cardiac care), it is possible that families with greater psychosocial risk are also less likely to experience strong partnerships with their cardiac care teams in a way that would support them in feeling comfortable engaging with outpatient medical care in the absence of an acute medical need. Families may feel apprehension about returning for preventive cardiology appointments if their child appears well, in the context of traumatic experiences with their child's cardiac care team during inpatient admissions.

Table 2. Multivariable Regressions

	β	*p*	Adjusted R^2	*SE*
Length of first surgical admission			.082	72.21
STAT score	.31	.008		
Total unplanned readmissions			.099	1.84
PAT Family Structure	.28	.005		
Diagnosis timing	-2.42	.02		
Total readmission days after first surgery			.324	34.10
Poverty	.41	<.001		
Single ventricle anatomy	.34	.001		
Return for neurodevelopmental follow-up			0.174	0.31
PAT Family Structure	-0.63	0.048		
Total CICU days	-0.01	0.011		
Diagnosis timing	-0.21	0.035		
Adherence to cardiology follow-up			0.084	7.34
PAT Total score	16.36	0.03		

CHAPTER 5: DISCUSSION

The current project aimed to examine both clinical and psychosocial factors that impact the cardiac care children with CHD receive in the first two years of life. Though research funding and hospital resources have flowed toward optimizing surgical and other clinical care techniques, results indicate that factors such as poverty and other SDH have a greater impact on many CHD outcomes. While medical factors were the strongest predictor of length of stay for a child's first surgical admission, psychosocial risk factors predicted number of unplanned readmissions, length of stay during these admissions, and likelihood of returning for recommended neurodevelopmental and cardiology outpatient follow-up.

Certainly this speaks to the degree to which family resources act as a barrier to cardiac care. However, it is important to consider that families who experience higher psychosocial risk may also be the ones who are not afforded the benefit of strong partnership with medical providers and high quality family centered care in the CICU, making them then less likely to seek regular preventive care with their child's outpatient cardiac team after hospital discharge. Families who experience discriminatory and neglectful care during hospital admission likely feel apprehensive about returning for outpatient care with the same providers when their child appears well. As suggested by the model, SDH and the quality of care families receive both interfere with the three pillars of cardiac care known to promote optimal medical and neurodevelopmental outcomes for children with CHD.

An urban bioethics approach may be particularly useful in looking toward next steps for addressing CHD outcome disparities. Wilmington is indeed a densely populated and highly segregated urban area, with a children's hospital that serves a racially, ethnically, socioeconomically, and linguistically diverse patient population, where sociodemographic factors are strong predictors of the care and care outcomes patients receive, and thus a bioethics lens is particularly well suited for examining the results of the current study and making broader recommendations to the field of CHD care.

Clinically, there is a gap between the care we know is most supportive of health outcomes and the care that all families are able to engage in and receive in CHD care. Supporting patient and family agency means engaging in power sharing practices through shared medical decision making as part of family centered care in the CICU, cardiac step-down unit, and outpatient settings. Cardiac providers must treat families as valued members of their child's care teams, demonstrating respect for their beliefs and perspectives, and engaging in regular reflexivity to identify and examine harmful biases.

Acting in solidarity with families means that CHD researchers must engage with diverse stakeholders most impacted by health disparities. Researchers must identify what Black and Hispanic patients and families perceive to be the most important research questions in CHD care and prioritize their perspectives on how to conduct meaningful research that does not put these communities at risk or place undue burden on their time, energy, and resources for the benefit of the researcher.

Social justice highlights the need for providers and researchers to also act as advocates in equitable reallocation of hospital and grant resources toward the patients at

greatest need. Rather than funding clinical research that targets factors unlikely to impact long-term, complex psychosocial outcomes, fund research that meaningfully impacts quality of life. Pay parents to sit at the bedside with their child. Pay parents to provide care to their child. Pay for parents to receive care for their own mental health. Research should be valued based on its scientific merit, not on volume or speed of production. Scientific merit in CHD outcomes research must be defined by its ability to address the disparities that have been well documented.

There is a need for multilevel interventions that emerge from the lived experiences and expertise of families of children with CHD experiencing the greatest disparities in health outcomes. With this gap in mind, I developed an interview guide to better understand the experiences of Black and Hispanic parents of children with CHD specifically, and to begin generating stakeholder driven recommendations to address disparities in care (Appendix A). This interview guide was reviewed and discussed by qualitative experts at the Lewis Katz School of Medicine's Center for Urban Bioethics, as well as members of the Cardiac Neurodevelopmental Outcome Collaborative, a group of multidisciplinary providers focused on improving neurodevelopmental outcomes for individuals with CHD. Next steps for this interview guide include partnering with families to further refine the interview guide, piloting it, collecting data, and moving forward toward the creation of an intervention specifically aimed at addressing health disparities for children with CHD.

We know that access to resources is a barrier to high quality cardiac care for children with CHD. An urban bioethics framework asks us to consider the ways in which

cardiac care teams are barriers to high quality care. To continue improving outcomes for children with CHD as a whole, the cardiac care community must divert resources, attention, and energy toward improving outcomes for those who have been historically marginalized and ignored by CHD research and the implementation of advancements in care techniques. Efforts to do this will be successful only if they are carried out with these patients and families centered as equal partners in research and clinical care.

WORKS CITED

Almli, L. M., Alter, C. C., Russell, R. B., Tinker, S. C., Howards, P. P., Cragan, J., Petersen, E., Carrino, G. E., & Reefhuis, J. (2017). Association Between Infant Mortality Attributable to Birth Defects and Payment Source for Delivery—United States, 2011–2013. *MMWR. Morbidity and Mortality Weekly Report*, *66*(3), 84–87. https://doi.org/10.15585/mmwr.mm6603a4

Bean Jaworski, J. L., Flynn, T., Burnham, N., Chittams, J. L., Sammarco, T., Gerdes, M., Bernbaum, J. C., Clancy, R. R., Solot, C. B., Zackai, E. H., McDonald-McGinn, D. M., & Gaynor, J. W. (2017). Rates of autism and potential risk factors in children with congenital heart defects. *Congenital Heart Disease*, *12*(4), 421–429. https://doi.org/10.1111/chd.12461

Beck, A. F., Edwards, E. M., Horbar, J. D., Howell, E. A., McCormick, M. C., & Pursley, D. M. (2020). The color of health: How racism, segregation, and inequality affect the health and well-being of preterm infants and their families. *Pediatric Research*, *87*(2), 227–234. https://doi.org/10.1038/s41390-019-0513-6

Blustein, J., & Fleischman, A. R. (2004). Urban bioethics: Adapting bioethics to the urban context. *Academic Medicine*, *79*(12), 1198–1202.

Bucholz, E. M., Sleeper, L. A., Goldberg, C. S., Pasquali, S. K., Anderson, B. R., Gaynor, J. W., Cnota, J. F., & Newburger, J. W. (2020). Socioeconomic Status and Long-term Outcomes in Single Ventricle Heart Disease. *Pediatrics*, *146*(4), e20201240. https://doi.org/10.1542/peds.2020-1240

Bucholz, E. M., Sleeper, L. A., Sananes, R., Brosig, C. L., Goldberg, C. S., Pasquali, S. K., & Newburger, J. W. (2021). Trajectories in Neurodevelopmental, Health-Related Quality of Life, and Functional Status Outcomes by Socioeconomic Status and Maternal Education in Children with Single Ventricle Heart Disease. *The Journal of Pediatrics*, *229*, 289-293.e3. https://doi.org/10.1016/j.jpeds.2020.09.066

Carbajal, R., Rousset, A., Danan, C., Coquery, S., Nolent, P., Ducrocq, S., Saizou, C., Lapillonne, A., Granier, M., Durand, P., Lenclen, R., Coursol, A., Hubert, P., Blanquat, L. de S., Boëlle, P.-Y., Annequin, D., Cimerman, P., Anand, K. J. S., & Bréart, G. (2008). Epidemiology and Treatment of Painful Procedures in Neonates in Intensive Care Units. *JAMA*, *300*(1), 60–70. https://doi.org/10.1001/jama.300.1.60

Daniels, J. M., & Harrison, T. M. (2016). A Case Study of the Environmental Experience of a Hospitalized Newborn Infant With Complex Congenital Heart Disease: *The Journal of Cardiovascular Nursing*, *31*(5), 390–398. https://doi.org/10.1097/JCN.0000000000000273

Davey, B., Sinha, R., Lee, J. H., Gauthier, M., & Flores, G. (2021). Social determinants of health and outcomes for children and adults with congenital heart disease: A systematic review. *Pediatric Research*, *89*(2), 275–294. https://doi.org/10.1038/s41390-020-01196-6

Egbe, A., Uppu, S., Stroustrup, A., Lee, S., Ho, D., & Srivastava, S. (2014). Incidences and Sociodemographics of Specific Congenital Heart Diseases in the United States of America: An Evaluation of Hospital Discharge Diagnoses. *Pediatric Cardiology*, *35*(6), 975–982. https://doi.org/10.1007/s00246-014-0884-8

Enlow, E., Faherty, L. J., Wallace-Keeshen, S., Martin, A. E., Shea, J. A., & Lorch, S. A. (2017). Perspectives of Low Socioeconomic Status Mothers of Premature Infants. *Pediatrics*, *139*(3), e20162310. https://doi.org/10.1542/peds.2016-2310

Fixler, D. E., Nembhard, W. N., Xu, P., Ethen, M. K., & Canfield, M. A. (2012). Effect of Acculturation and Distance From Cardiac Center on Congenital Heart Disease Mortality. *PEDIATRICS*, *129*(6), 1118–1124. https://doi.org/10.1542/peds.2011-3114

Gilboa, S. M., Salemi, J. L., Nembhard, W. N., Fixler, D. E., & Correa, A. (2010). Mortality Resulting From Congenital Heart Disease Among Children and Adults in the United States, 1999 to 2006. *Circulation*, *122*(22), 2254–2263. https://doi.org/10.1161/CIRCULATIONAHA.110.947002

Goldberg, C. S., Hu, C., Brosig, C., Gaynor, J. W., Mahle, W. T., Miller, T., Mussatto, K. A., Sananes, R., Uzark, K., Trachtenberg, F., Pizarro, C., Pemberton, V. L., Lewis, A. B., Li, J. S., Jacobs, J. P., Cnota, J., Atz, A. M., Lai, W. W., Bellinger, D., … PHN INVESTIGATORS. (2019). Behavior and Quality of Life at 6 Years for Children With Hypoplastic Left Heart Syndrome. *Pediatrics*, *144*(5), e20191010. https://doi.org/10.1542/peds.2019-1010

Harrison, T. M., & Brown, R. (2017). Autonomic Nervous System Function Following a Skin-to-Skin Contact Intervention in Infants with Congenital Heart Disease. *The Journal of Cardiovascular Nursing*, *32*(5), E1–E13. https://doi.org/10.1097/JCN.0000000000000397

Hickey, P. A., Connor, J. A., Cherian, K. M., Jenkins, K., Doherty, K., Zhang, H., Gaies, M., Pasquali, S., Tabbutt, S., St. Louis, J. D., Sarris, G. E., Kurosawa, H., Jonas, R. A., Sandoval, N., Tchervenkov, C. I., Jacobs, J. P., Stellin, G., Kirklin, J. K., Garg, R., & Vener, D. F. (2017). International quality improvement initiatives. *Cardiology in the Young*, *27*(S6), S61–S68. https://doi.org/10.1017/S1047951117002633

Hill, G., Block, J., Tanem, J., & Frommelt, M. (2015). Disparities in the Prenatal Detection of Critical Congenital Heart Disease. *Prenatal Diagnosis*, *35*(9), 859–863. https://doi.org/10.1002/pd.4622

Howell, E. A. (2008). Racial Disparities in Infant Mortality: A Quality of Care Perspective. *Mount Sinai Journal of Medicine*, *75*(1), 31–35. https://doi.org/10.1002/msj.20018

Jacobs, J. P. (2015). The science of assessing the outcomes and improving the quality of the congenital and paediatric cardiac care. *Current Opinion in Cardiology*, *30*(1), 100–111.

Kazak, A. E., Schneider, S., Didonato, S., & Pai, A. L. H. (2015). Family psychosocial risk screening guided by the Pediatric Psychosocial Preventative Health Model (PPPHM) using the Psychosocial Assessment Tool (PAT). *Acta Oncologica*, *54*(5), 574–580. https://doi.org/10.3109/0284186X.2014.995774

Kilbourne, A. M., Switzer, G., Hyman, K., Crowley-Matoka, M., & Fine, M. J. (2006). Advancing Health Disparities Research Within the Health Care System: A Conceptual Framework. *American Journal of Public Health*, *96*(12), 2113–2121. https://doi.org/10.2105/AJPH.2005.077628

Kucik, J. E., Cassell, C. H., Alverson, C. J., Donohue, P., Tanner, J. P., Minkovitz, C. S., Correia, J., Burke, T., & Kirby, R. S. (2014). Role of Health Insurance on the Survival of Infants With Congenital Heart Defects. *American Journal of Public Health*, *104*(9), e62–e70. https://doi.org/10.2105/AJPH.2014.301969

Kucik, J. E., Nembhard, W. N., Donohue, P., Devine, O., Wang, Y., Minkovitz, C. S., & Burke, T. (2014). Community Socioeconomic Disadvantage and the Survival of Infants With Congenital Heart Defects. *American Journal of Public Health*, *104*(11), e150–e157. https://doi.org/10.2105/AJPH.2014.302099

Laing, S., McMahon, C., Ungerer, J., Taylor, A., Badawi, N., & Spence, K. (2010). Mother–child interaction and child developmental capacities in toddlers with major birth defects requiring newborn surgery. *Early Human Development*, *86*(12), 793–800. https://doi.org/10.1016/j.earlhumdev.2010.08.025

Lee, J. S., Cinanni, N., Di Cristofaro, N., Lee, S., Dillenburg, R., Adamo, K. B., Mondal, T., Barrowman, N., Shanmugam, G., Timmons, B. W., & Longmuir, P. W. (2020). Parents of Very Young Children with Congenital Heart Defects Report Good Quality of Life for Their Children and Families Regardless of Defect Severity. *Pediatric Cardiology*, *41*(1), 46–53. https://doi.org/10.1007/s00246-019-02220-1

Lewis, T. P., Andrews, K. G., Shenberger, E., Betancourt, T. S., Fink, G., Pereira, S., & McConnell, M. (2019). Caregiving can be costly: A qualitative study of barriers and facilitators to conducting kangaroo mother care in a US tertiary hospital neonatal intensive care unit. *BMC Pregnancy and Childbirth*, *19*(1), 227. https://doi.org/10.1186/s12884-019-2363-y

Ligsay, A., & Goldberg, C. S. (2021). An introduction to and review of cardiac neurodevelopment: The risks and recommended approaches. *Current Opinion in Pediatrics*, *33*(5), 489–494. https://doi.org/10.1097/MOP.0000000000001057

Lisanti, A. J. (2018). Parental stress and resilience in CHD: A new frontier for health disparities research. *Cardiology in the Young*, *28*(9), 1142–1150. https://doi.org/10.1017/S1047951118000963

Lisanti, A. J., Allen, L. R., Kelly, L., & Medoff-Cooper, B. (2017). Maternal Stress and Anxiety in the Pediatric Cardiac Intensive Care Unit. *American Journal of Critical Care : An Official Publication, American Association of Critical-Care Nurses*, *26*(2), 118–125. https://doi.org/10.4037/ajcc2017266

Lisanti, A. J., Vittner, D., Medoff-Cooper, B., Fogel, J., Wernovsky, G., & Butler, S. (2019). Individualized Family-Centered Developmental Care: An Essential Model to Address the Unique Needs of Infants With Congenital Heart Disease. *The Journal of Cardiovascular Nursing*, *34*(1), 85–93. https://doi.org/10.1097/JCN.0000000000000546

Lopez, K. N., Morris, S. A., Tejtel, K. S., Espaillat, A., & Salemi, J. L. (2020). *US Mortality Due To Congenital Heart Disease Across the Lifespan from 1999-2017 Exposes Persistent Racial/Ethnic Disparities* [Preprint]. Cardiovascular Medicine. https://doi.org/10.1101/2020.03.15.20036525

Lu, J. C. . ., Lowery, R., Yu, S., Ghadimi Mahani, M., Agarwal, P. P., & Dorfman, A. L. (2017). Predictors of missed appointments in patients referred for congenital or pediatric cardiac magnetic resonance. *Pediatric Radiology*, *47*(8), 911–916. https://doi.org/10.1007/s00247-017-3851-8

Lushaj, E. B., Hermsen, J., Leverson, G., MacLellan-Tobert, S. G., Nelson, K., Amond, K., & Anagnostopoulos, P. V. (2020). Beyond 30 Days: Analysis of Unplanned Readmissions During the First Year Following Congenital Heart Surgery. *World Journal for Pediatric and Congenital Heart Surgery*, *11*(2), 177–182. https://doi.org/10.1177/2150135119895212

Mackie, A. S., Rempel, G. R., Rankin, K. N., Nicholas, D., & Magill-Evans, J. (2012). Risk factors for loss to follow-up among children and young adults with congenital heart disease. *Cardiology in the Young*, *22*(3), 307–315. https://doi.org/10.1017/S104795111100148X

Majnemer, A., Limperopoulos, C., Shevell, M., Rohlicek, C., Rosenblatt, B., & Tchervenkov, C. (2008). Developmental and Functional Outcomes at School Entry in Children with Congenital Heart Defects. *The Journal of Pediatrics*, *153*(1), 55-60.e1. https://doi.org/10.1016/j.jpeds.2007.12.019

Marino, B. S., Lipkin, P. H., Newburger, J. W., Peacock, G., Gerdes, M., Gaynor, J. W., Mussatto, K. A., Uzark, K., Goldberg, C. S., Johnson, W. H., Li, J., Smith, S. E., Bellinger, D. C., & Mahle, W. T. (2012). Neurodevelopmental Outcomes in Children With Congenital Heart Disease: Evaluation and Management: A Scientific Statement From the American Heart Association. *Circulation*, *126*(9), 1143–1172. https://doi.org/10.1161/CIR.0b013e318265ee8a

McCusker, C. G., Doherty, N. N., Molloy, B., Casey, F., Rooney, N., Mulholland, C., Sands, A., Craig, B., & Stewart, M. (2007). Determinants of neuropsychological and behavioural outcomes in early childhood survivors of congenital heart disease. *Archives of Disease in Childhood*, *92*(2), 137–141.

Mulkey, S. B., Bai, S., Luo, C., Cleavenger, J. E., Gibson, N., Holland, G., Mosley, B. S., Kaiser, J. R., & Bhutta, A. T. (2016). School-Age Test Proficiency and Special Education after Congenital Heart Disease Surgery in Infancy. *The Journal of Pediatrics*, *178*, 47-54.e1. https://doi.org/10.1016/j.jpeds.2016.06.063

Mulkey, S. B., & du Plessis, A. J. (2019). Autonomic Nervous System Development and its' Impact on Neuropsychiatric Outcome. *Pediatric Research*, *85*(2), 120–126. https://doi.org/10.1038/s41390-018-0155-0

Pace, N. D., Oster, M. E., Forestieri, N. E., Enright, D., Knight, J., & Meyer, R. E. (2018). *Sociodemographic Factors and Survival of Infants With Congenital Heart Defects*. *142*(3), 13.

Peiris, V., Singh, T. P., Tworetzky, W., Chong, E. C., Gauvreau, K., & Brown, D. W. (2009). Association of Socioeconomic Position and Medical Insurance With Fetal Diagnosis of Critical Congenital Heart Disease. *Circulation: Cardiovascular Quality and Outcomes*, *2*(4), 354–360. https://doi.org/10.1161/CIRCOUTCOMES.108.802868

Peterson, J. K., Chen, Y., Nguyen, D. V., & Setty, S. P. (2017). Current trends in racial, ethnic, and healthcare disparities associated with pediatric cardiac surgery outcomes. *Congenital Heart Disease*, *12*(4), 520–532. https://doi.org/10.1111/chd.12475

Peyvandi, S., Baer, R. J., Moon-Grady, A. J., Oltman, S. P., Chambers, C. D., Norton, M. E., Rajagopal, S., Ryckman, K. K., Jelliffe-Pawlowski, L. L., & Steurer, M. A. (2018). Socioeconomic Mediators of Racial and Ethnic Disparities in Congenital Heart Disease Outcomes: A Population-Based Study in California. *Journal of the American Heart Association*, *7*(20). https://doi.org/10.1161/JAHA.118.010342

Phillips, R. M. (2015). Seven Core Measures of Neuroprotective Family-Centered Developmental Care: Creating an Infrastructure for Implementation. *Newborn and Infant Nursing Reviews*, *15*(3), 87–90. https://doi.org/10.1053/j.nainr.2015.06.004

Profit, J., Gould, J. B., Bennett, M., Goldstein, B. A., Draper, D., Phibbs, C. S., & Lee, H. C. (2017). Racial/Ethnic Disparity in NICU Quality of Care Delivery. *Pediatrics*, *140*(3). https://doi.org/10.1542/peds.2017-0918

Purkey, N. J., Axelrod, D. M., McElhinney, D. B., Rigdon, J., Qin, F., Desai, M., Shin, A. Y., Chock, V. Y., & Lee, H. C. (2019). Birth Location of Infants with Critical Congenital Heart Disease in California. *Pediatric Cardiology*, *40*(2), 310–318. https://doi.org/10.1007/s00246-018-2019-0

Reller, M. D., Strickland, M. J., Riehle-Colarusso, T., Mahle, W. T., & Correa, A. (2008). Prevalence of Congenital Heart Defects in Metropolitan Atlanta, 1998–2005. *The Journal of Pediatrics*, *153*(6), 807–813. https://doi.org/10.1016/j.jpeds.2008.05.059

Ryan, K. R., Jones, M. B., Allen, K. Y., Marino, B. S., Casey, F., Wernovsky, G., & Lisanti, A. J. (2019). Neurodevelopmental Outcomes Among Children With Congenital Heart Disease: At-Risk Populations and Modifiable Risk Factors. *World Journal for Pediatric and Congenital Heart Surgery*, *10*(6), 750–758. https://doi.org/10.1177/2150135119878702

Siffel, C., Riehle-Colarusso, T., Oster, M. E., & Correa, A. (2015). Survival of Children With Hypoplastic Left Heart Syndrome. *Pediatrics*, *136*(4), e864–e870. https://doi.org/10.1542/peds.2014-1427

Sigurdson, K., Morton, C., Mitchell, B., & Profit, J. (2018). Disparities in NICU quality of care: A qualitative study of family and clinician accounts. *Journal of Perinatology*, *38*(5), 600–607. https://doi.org/10.1038/s41372-018-0057-3

Sigurdson, K., Profit, J., Dhurjati, R., Morton, C., Scala, M., Vernon, L., Randolph, A., Phan, J. T., & Franck, L. S. (2020). Former NICU Families Describe Gaps in Family-Centered Care. *Qualitative Health Research*, *30*(12), 1861–1875. https://doi.org/10.1177/1049732320932897

Sood, E., Karpyn, A., Demianczyk, A. C., Ryan, J., Delaplane, E. A., Neely, T., Frazier, A. H., & Kazak, A. E. (2018). Mothers and Fathers Experience Stress of Congenital Heart Disease Differently: Recommendations for Pediatric Critical Care. *Pediatric Critical Care Medicine*, 1. https://doi.org/10.1097/PCC.0000000000001528

Sood, E., Wysocki, T., Alderfer, M. A., Aroian, K., Christofferson, J., Karpyn, A., Kazak, A. E., & Pierce, J. (2021). Topical Review: Crowdsourcing as a Novel Approach to Qualitative Research. *Journal of Pediatric Psychology*, *46*(2), 189–196. https://doi.org/10.1093/jpepsy/jsaa096

Tashiro, J., Wang, B., Sola, J. E., Hogan, A. R., Neville, H. L., & Perez, E. A. (2014). Patent ductus arteriosus ligation in premature infants in the United States. *Journal of Surgical Research*, *190*(2), 613–622. https://doi.org/10.1016/j.jss.2014.02.003

US Department of Health and Human Services. (2010). An opportunity to address societal determinants of health in the US July 26, 2010. *US Department of Health and Human Services, Office of Disease Prevention and Health Promotion. Https://Www. Healthypeople. Gov/Sites/Default/Files/SocietalDeterminantsHealth. Pdf*.

van der Linde, D., Konings, E. E. M., Slager, M. A., Witsenburg, M., Helbing, W. A., Takkenberg, J. J. M., & Roos-Hesselink, J. W. (2011). Birth Prevalence of Congenital Heart Disease Worldwide. *Journal of the American College of Cardiology*, *58*(21), 2241–2247. https://doi.org/10.1016/j.jacc.2011.08.025

Verrall, C. E., Blue, G. M., Loughran-Fowlds, A., Kasparian, N., Gecz, J., Walker, K., Dunwoodie, S. L., Cordina, R., Sholler, G., Badawi, N., & Winlaw, D. (2019). 'Big issues' in neurodevelopment for children and adults with congenital heart disease. *Open Heart*, *6*(2), e000998. https://doi.org/10.1136/openhrt-2018-000998

Wernovsky, G., & Licht, D. J. (2016). Neurodevelopmental Outcomes in Children With Congenital Heart Disease—What Can We Impact?: *Pediatric Critical Care Medicine*, *17*, S232–S242. https://doi.org/10.1097/PCC.0000000000000800

Woolf-King, S. E., Anger, A., Arnold, E. A., Weiss, S. J., & Teitel, D. (2017). Mental Health Among Parents of Children With Critical Congenital Heart Defects: A Systematic Review. *Journal of the American Heart Association*, *6*(2). https://doi.org/10.1161/JAHA.116.004862

Wren, C., & O'Sullivan, J. J. (2013). Loss to follow-up of adults with repaired congenital heart disease. *Heart*, *99*(7), 440–441. https://doi.org/10.1136/heartjnl-2012-303121

Wu, Y., Kapse, K., Jacobs, M., Niforatos-Andescavage, N., Donofrio, M. T., Krishnan, A., Vezina, G., Wessel, D., Plessis, A. du, & Limperopoulos, C. (2020). Association of Maternal Psychological Distress With In Utero Brain Development in Fetuses With Congenital Heart Disease. *JAMA Pediatrics*, *174*(3), e195316–e195316. https://doi.org/10.1001/jamapediatrics.2019.5316

APPENDIX A: INTERVIEW GUIDE

Introductory script: Thank you for agreeing to talk with me today. I am interested in hearing about the experiences of families who have a child with congenital heart disease. I am specifically interested in hearing about how cardiology care might be provided or experienced differently for families of different racial, ethnic, and cultural backgrounds. The information you share with me today will help us to provide better care for families like yours in the future.

I will be asking you about some topics that might be difficult to discuss. It is important for you to know that anything you share with me will not be shared with your child's nurses or doctors. No one else will know that you participated in this study or what you shared. I will be recording this interview and taking notes as we talk, but you and your child's names will not be linked to anything you say. You do not have to tell me anything you do not want to. Before we get started, what questions do you have?

1. Tell me about your child.
    1. How did you first learn about your child's CHD diagnosis?
2. I'd like to ask you about your experiences when your child was hospitalized in the intensive care unit after open heart surgery.
    1. When thinking back to this time, what was your relationship like with your child's doctors and nurses?
        1. Can you tell me about a time when you felt really connected to your child's medical team?
        2. Other parents have told me that they did not feel respected by their child's medical team or were not included in the decisions their child's team made about their care. Can you tell me about a time when you felt judged, neglected, or discriminated against by your child's medical team?
            1. *If yes follow up with*: Why do you think you were judged/neglected/discriminated against?
            2. *If indicated follow up with*: Do you think your race/ethnicity played a role?
        3. Can you describe your relationship with the medical team in a word?
    2. What made it more difficult for you to be at your child's bedside during hospitalization?
        1. *Probe for barriers including transportation, work/income, child care, access to meals/housing*
    3. What made it easier to be at the hospital?
        1. *Probe for facilitators including social work support*
    4. How could your child's cardiac center have improved their care of your child when he/she was hospitalized after open heart surgery?
3. Now I'd like to ask you about your experiences with your child's medical care after he/she was discharged from the hospital. What kind of follow-up care did your child need? For example, follow-up cardiology appointments, EI/therapies, educational services or supports (*tailor as appropriate depending on child s age)*.
    1. Can you tell me about a time when someone from your child's care team helped you access the services your child needed?

2. What made it more difficult to access the care your child's doctors recommended or the care you believe your child needs?
    1. *Probe for barriers including transportation, work/income, child-care, communication difficulties with care team*
    2. Some parents have told me that they did not feel supported by their child's treatment team in getting the care their child needed. When thinking about your child's long-term follow-up care, can you tell me about a time when you felt judged, neglected, or discriminated against by your child's cardiac care team? For example, your child's outpatient cardiologist, clinic staff, or therapists?
        1. *If yes follow up with*: Why do you think you were judged/neglected/discriminated against?
        2. *If indicated follow up with*: Do you think your race/ethnicity played a role?
3. How could your child's cardiac center have improved your child's follow-up care or your access to this care?