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The Relationships Between Parenting Stress, Growth, and Development in Infants with Congenital Heart Defects During the First Six Months of Life

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Abstract

The stress experienced by parents at the time of diagnosis and hospitalization for their infant's congenital heart defect (CHD) is well recognized by healthcare professionals. Increased parenting stress has been negatively correlated with development in low birthweight infants. The primary purpose of this study was to explore the parenting stress as experienced by parents of infants with CHD during their first six months of life. In addition, the relationship between parenting stress and the growth and development of infants with CHD was explored. Due to the transactional nature of mother-infant interaction, both directions of this relationship were examined, the factors of parenting stress predictive of growth and development and the factors of growth and development predictive of parenting stress. The change in stress over time was also evaluated. From a larger parent study examining feeding and energy balance in infants with CHD during their first year of life, 60 mother-infant dyads with complete data were selected. Thirty-five of these infants had a CHD (11 with single ventricle [SV] physiology) and 25 were healthy controls. Mothers completed infant temperament questionnaires and the Parenting Stress Index at 3 and 6 months, growth was also measured at these time points, and development was measured at 6 months utilizing the Bayley Scales of Infant Development-II. There were marked differences between subjects and controls; however, infants with SV physiology were found to bear an unequal share of adverse outcomes for infant temperament, parenting stress, and growth. Parenting stress correlated with and predicted growth and development. Growth and development however, did not predict parenting stress. It was predicted by temperament characteristics that comprise the "difficult" child constellation. Parenting stress decreased over time for all three groups. These original findings support the incorporation of parenting stress as a psychosocial variable in the exploration of biological phenomena such as infant growth and development. The importance of anticipatory guidance for parents of infants with SV physiology is stressed as well as the continued investigation of dyads to determine if the reported relationships in the first 6 months existed throughout the first year of life.

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THE RELATIONSHIPS BETWEEN PARENTING STRESS, GROWTH, AND DEVELOPMENT IN INFANTS WITH CONGENITAL HEART DEFECTS DURING THE FIRST SIX MONTHS OF LIFE

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Dedication

To Brock, for lovingly teaching me more about parenting than any dissertation ever could. It is such a joy to be your mommy.

To Ben, for being such a patient and supportive husband during this entire process and filling in the mommy gaps when I couldn't.

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"In all thy ways acknowledge him, and he will direct your paths." (Proverbs 3:6)

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ABSTRACT

THE RELATIONSHIPS BETWEEN PARENTING STRESS, GROWTH, AND DEVELOPMENT IN INFANTS WITH CONGENITAL HEART DEFECTS DURING THE FIRST SIX MONTHS OF LIFE

Danica Fulbright Sumpter, CRNP, MSN

Barbara Medoff-Cooper, RN, PhD, FAAN

The stress experienced by parents at the time of diagnosis and hospitalization for their infant's congenital heart defect (CHD) is well recognized by healthcare professionals. Increased parenting stress has been negatively correlated with development in low birthweight infants. The primary purpose of this study was to explore the parenting stress as experienced by parents of infants with CHD during their first six months of life. In addition, the relationship between parenting stress and the growth and development of infants with CHD was explored. Due to the transactional nature of mother-infant interaction, both directions of this relationship were examined, the factors of parenting stress predictive of growth and development and the factors of growth and development predictive of parenting stress. The change in stress over time was also evaluated. From a larger parent study examining feeding and energy balance in infants with CHD during their first year of life, 60 mother-infant dyads with complete data were selected. Thirty-five of these infants had a CHD (11 with single ventricle [SV] physiology) and 25 were healthy controls. Mothers completed infant temperament questionnaires and the Parenting Stress Index at 3 and 6 months, growth was also measured at these time points, and development was measured at 6 months utilizing the Bayley Scales of Infant Development-II. There were marked differences between

subjects and controls; however, infants with SV physiology were found to bear an unequal share of adverse outcomes for infant temperament, parenting stress, and growth. Parenting stress correlated with and predicted growth and development. Growth and development however, did not predict parenting stress. It was predicted by temperament characteristics that comprise the "difficult" child constellation. Parenting stress decreased over time for all three groups. These original findings support the incorporation of parenting stress as a psychosocial variable in the exploration of biological phenomena such as infant growth and development. The importance of anticipatory guidance for parents of infants with SV physiology is stressed as well as the continued investigation of dyads to determine if the reported relationships in the first 6 months existed throughout the first year of life.

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Chapter 1: THE PROBLEM

Introduction

Becoming a parent, one of the most powerful of the human experiences, is often accompanied with feelings of celebration and relief, but it can also be a time of anxiety, and stress (Lawoko & Soares, 2002). The term "parenting" is derived from the Latin root *pario*, meaning life-giver, and encompasses much more than just the caregiving activities parents perform (Scher & Sharabany, 2005). Parenting frequently involves pleasure and joy and provides individuals with a sense of competence, but at times parenting can be confusing, frustrating, irritating, and stressful-even with a "healthy" child (Scher & Sharabany, 2005). One can only imagine the increase in stress that takes place when the hopes and dreams of the "perfect" pregnancy, labor, and delivery are shattered with the revelation of a congenital heart defect (CHD), and the grieving process that ensues as parents cope with the challenges of having an infant with CHD.

Statement of the Problem

Parental stress, or the stress produced by parenting, arises from different sources such as the severity of the infant's illness, the infant's temperament, various sociodemographic factors, and delays in physical growth and cognitive development. The stress experienced by parents at the time of their infant's CHD diagnosis and/or hospitalization is well recognized by healthcare professionals (Svavarsdottir & McCubbin, 1996; Goldberg et al, 1997; Visconti, 2002). The birth of a healthy infant is stressful in and of itself (Willinger, 2005). Adding to the uncertainty of a potentially life-threatening diagnosis of CHD, there are often imperative decisions to be made about

open heart surgery within the first months of life, along with the grief experienced from losing their (concept of a) "perfect" infant, and it is clear why parents of infants with CHD report heightened levels of stress compared to parents of healthy infants (Miles & Clark, 1999; Visconti et al, 2002). This stress is compounded by the intensive care environment, the complexity of caring for these infants, and lastly, the infant him/herself (i.e. severity of the infant's CHD and infant temperament).

The intensive care environment is fraught with stress-inducing sights and sounds and multiple factors decreasing parents' ability to "parent" (e.g. comfort/hold a crying infant), but the most obvious factor influencing parenting stress during hospitalization involves the infant him/herself (Dudek-Shriber, 2004). The appearance of the infant and the inability to establish and maintain typical parent-child interactions adds to the stress these parents may be experiencing. In addition, the possibility of transport that separates infants from parents and/or the need for surgery within the first few days of life can compound the situation. Unfortunately, this stress is not confined to the inpatient facility. The physical condition of the infant, such as severity of the CHD and other co-morbidities and syndromes often necessitate complex caregiving after discharge, which can cause the stress these parents experience to persist and increase even after discharge from the hospital. Parenting a chronically ill child creates increased levels of stress in families who possess access to medical, social, and financial resources; understandably, families without access or with limited access to these types of supports have additional challenges and subsequent increased levels of stress (Browne & Talmni, 2005).

Not only does the physical condition and appearance of the infant impact stress, but the behavioral style or specifically the infant's temperament can contribute to parenting

stress as well. Infants with "difficult" temperament characteristics have been found to present a greater challenge for parents (Secco & Moffatt, 2003). Parental distress significantly increases the likelihood of an infant being perceived as difficult (Mäntymaa, Mirjami et al, 2006; Sheinkopf, 2006), and this type of temperament also contributes negatively to parenting competence and increases parenting stress (Secco & Moffatt, 2003; Gutteling 2005). This provides support for a cyclical or bi-directional relationship between infant characteristics (e.g. illness and temperament) and parenting stress.

Definition of Terms

Parenting stress or the stress generated by parenting is conceptualized by Richard Abidin, author of the Parenting Stress Index (PSI), as being comprised of salient parental characteristics, child characteristics, and situational variables directly related to the parental role (Abidin, 1995). His model postulates that the stressors a parent experiences related to the role of being a parent will influence parenting behavior, which can in turn have an impact on the psychosocial adaptation of the child.

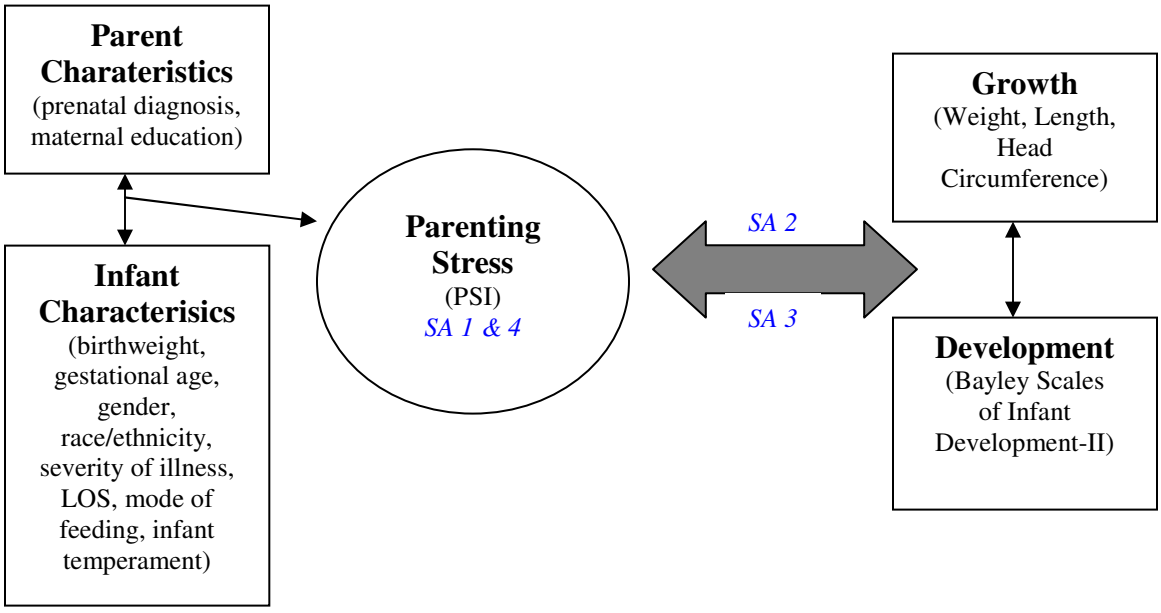
Parent characteristics include maternal education, and whether or not the CHD was diagnosed pre- or postnatally. *Infant characteristics* include severity of illness, which captures the complexity of an infant's heart defect and resulting surgical repair. This variable is operationalized using the Risk Adjustment in Congenital Heart Surgery-1 (RACHS-1) scoring system which uses surgical procedures as the primary proxy for in-hospital mortality risk. Other infant variables include gestational age, birthweight, gender, race/ethnicity, and infant temperament. Infant temperament is the inborn behavioral style of every infant. It is measured by the Early Infant Temperament Questionnaire (EITQ) as well as the Infant Temperament Questionnaire (ITQ). These

instruments measure nine temperament characteristics and based on objective and subjective parental ratings infants are classified as “easy”, “slow to warm up”, or “difficult”. The “difficult” temperament constellation is directly associated with increased parenting stress.

Growth as defined in this study is physical growth operationalized using the anthropometric measurements of weight, length, and head circumference.

Development is defined as the process of cognitive maturation and is measured by the Bayley Scales of Infant Development- II. This study specifically examines the Mental Development Index (MDI) and the Psychomotor Development Index (PDI).

Theoretical Framework



Study Purpose & Specific Aims

The purpose of this study is to explore the parenting stress as experienced by parents of infants with CHD during the first six months of life and the relationship between this stress and the growth and development of these infants. This will be achieved by addressing four specific aims:

1.) Describe the parenting stress experienced by parents of infants with CHD at three and six months of life

2.) Examine factors of parenting stress which are associated with and predict growth and development

3.) Examine factors of growth and development which are associated with and predict parenting stress

4.) Identify the changes in parenting stress over the course of the first six months of life

Significance

Stress is a major concern for families of infants with CHD and may have implications for the physical growth and cognitive development of these infants. Increased parenting stress has been negatively correlated with development in low birth weight infants (Robson, 1997), but it is not known what effects parenting stress has on growth or development in infants with CHD. The proposed study will provide an understanding of parenting stress and how it relates to and effects the physical growth and cognitive development of infants with CHD. Parenting stress is a mutable variable

and one amenable to interventions with parents as well as healthcare providers to reduce stress levels and improve parenting and coping skills. This study will provide valuable foundational information in this developing area of research.

Chapter 2: BACKGROUND LITERATURE

Parenting stress

The stress experienced by parents at the time of their infant's congenital heart defect (CHD) diagnosis or hospitalization is well recognized by healthcare professionals (Svavarsdottir & McCubbin, 1996; Goldberg et al, 1997; Visconti et al, 2002). Numerous factors influence a person's ability to parent a child with CHD and other chronic illnesses. The health status of a child is one of many factors that can contribute to the parental stress which may in turn influence the quality of child rearing (Carey, Nicholson, Fox, 2002). The presence of a chronic illness may create an even more challenging future than parents anticipated and how they respond can affect both the short and long term developmental outcomes for their children (Carey, Nicholson, Fox, 2002).

Parents of infants with CHD have identified psychological stress as one of their most significant problems, (Green, 2004) and during the infant's first year of life, feeding has long been one of the most stressful situations for these families (D'Antonio, 1979). In a survey of caregiving tasks by Svavarsdottir and McCubbin (1996), feeding proved to be the most time consuming and third most difficult task for mothers (behind providing emotional support for spouse/partner and managing discipline and behavior problems such as crying and irritability). This is possibly due to the shortness of breath, cyanosis, the mother's insecurity in reading the infant's cues that can occur during feeding, or her lack of information on how to handle the infant during feeding (Svavarsdottir & McCubbin, 1996). Lobo found that infants with CHD responded less to their mothers during feeding than healthy infants and their mothers provided less social/emotional, growth fostering opportunities for their infants during feeding (Lobo, 1992). This meant

that mothers of infants with CHD were less apt to smile, make eye contact, touch, hum, or sing during feeding than mothers of control infants. Lobo gives two possible explanations for this. It could be that infants with CHD present behaviors at birth that disrupt the feeding or it may be that the infant develops behaviors difficult for the mother to interpret over time. The anxiety a mother experiences due to the knowledge that her infant could choke and/or die during a feeding leads her to feed differently, or it might be that the mother has noticed when she performs the social/emotional growth fostering activities the infant eats less (Lobo, 1992). If the infants with CHD have learned to moderate the amount of stimulation they absorb from the environment during feeding, they may appear less responsive to the caregiver which may in turn frustrate the caregiver and lead him/her to be less responsive to the infant as well (Lobo, 1992).

Feeding is much more than just a means of obtaining nutrients. It is an activity with many social and cultural implications (Imms, 2001). Oral infant feeding is also one of the expectations of a "normal" infant, and when an infant with CHD feeds by mouth successfully, this can have a normalizing effect for parents. The feeding problems that can occur in this population include a dyscoordination of sucking, swallowing, and breathing; inefficient and weak suck; emesis and gastro-esophageal reflux (Pillo-Blocka, Adatia, Sharieff, McCrindle, & Zlotkin, 2004; Steltzer et al., 2005).

Intimately related to the feeding issue is the concern over physical growth. Infants needing multiple surgeries often need to gain a specific amount of weight before the surgery can be performed. This can cause an additional stressor as the healthcare team works along with parents to increase caloric intake and minimize energy expenditure. Weight gain is also an important signal to parents that their infant is growing and

developing “normally”, and because growth is the single most important parameter in assessing the health status of an infant, it is equally important to health care providers as well (Lipman et al, 2004). When infants do not gain weight at age and gender appropriate rates, this can be disappointing and stressful to parents, especially when they are currently doing all that has been prescribed (e.g. increased caloric density feedings via nasogastric or gastronomy tubes, etc.). Despite the efforts to increase calories and decrease energy expenditure, as many as 50% of infants with CHD are malnourished and receive a diagnosis of failure to thrive (FTT) (Peterson & Wetzel, 2004; Avitzur et al., 2003). The malnutrition that occurs not only affects the physical growth of these infants, but their cognitive development as well, which can lead to another potential source of parenting stress (Chi-Wen Chen, Chung-Yi Li, & Jou-Kou Wang, 2004; Nydegger & Bines, 2006; Miles, Carter, Hennessey, Eberly, & Riddle, 1989).

Along with feeding and infant growth concerns, environmental factors associated with parenting stress include the hospital intensive care unit (ICU) environment (and subsequent alterations in the parent-infant relationship), available resources, and other life stresses. Though the cardiac ICU environment has not been examined to date, the pediatric and neonatal ICU environments serve as comparable comparisons. The most stressful aspects of the pediatric intensive care unit (PICU) experience (17% of these children had CHD) were found to be related to the child’s behavior and emotions and regarding parental role alteration. The items from the Parental Stressor Scale: PICU (PSS: PICU) subscale, “Child’s Behavior and Emotion”, that were most stressful were, “seeing my child in pain”, “seeing the child frightened and sad”, and the “inability of the child to communicate with the parent” (Miles, Carter, Riddle, Hennessey, & Eberly,

1989). The items from the "Parent Role Alteration" dimension of the PSS: PICU with the highest stress scores were, "feeling unable to protect my child" and "not knowing how to best help my child". These findings suggest that alterations in the parent-child relationship may indeed be more stressful than the actual ICU environment itself, and these feelings of helplessness in the parental role are a great source of stress for parents and a potential area of intervention for nurses (Miles et al., 1989).

Specific sources of stress were examined in the neonatal intensive care unit (NICU) by using self-report measures of acute stress and parent perceptions of stress in the NICU. It was found that alteration of parent role, which included not being able to help, hold, or care for the infant, protect the infant from pain, or share the infant with other family members were the factors most strongly associated with symptoms of acute stress disorder (Shaw et al., 2006). Acute stress disorder (ASD) is the form of traumatic stress experienced in the first weeks after a traumatic event; it is considered to be a precursor to post-traumatic stress disorder (PTSD) (Shaw et al., 2006).

Though it is difficult to determine sequence and causality, families with more stress have also reported fewer resources (Visconti et al., 2002). In examining distress and hopelessness, it was found that variables such as employment status and financial situation explained more of the variance than did the disease process itself (CHD and other diseases) (Lawoko & Soares, 2002). Parental role strain has been shown to increase when there are two or more children in the home, parents are employed, in mothers (especially single mothers), and in lower income families (Vilhjalmsson & Kristjansdottir, 2006). Because people do not live in silos, it is important to take these

environmental factors into consideration when assessing potential sources of parent stress.

Stress, though not inherently good or bad, is necessary in moderate amounts in daily life to stimulate optimum performance, but when parenting stress is sustained without effective coping mechanisms to address it, negative effects for both parents and children can ensue. Goldberg et al. have reported that parenting stress over the first three years of children's lives is the best predictor of child behavior problems at four years of age (1997). These child behavior problems (e.g. internalizing and externalizing behaviors) are associated with higher parent stress, and negatively correlated with the psychosocial well-being of the child (Majnemer et al., 2006). Positive parent-infant attachment, which may be altered by parenting stress, is necessary for fostering the optimal growth and development of an infant and for encouraging the nascent parent-infant relationship (Schenk, Kelley, & Schenk, 2005). Additionally, securely attached infants with CHD have demonstrated greater improvements in their physical health than those less securely attached infants (Carey et al., 2002).

Parenting stress not only affects the parent-infant relationship directly, but it also affects the parents' mental and physical health, which subsequently affects the infant as well. The formation of depressive symptoms can lead to decreased parental responsiveness and sensitivity to infants cues (Swartz, 2005). This decreased responsiveness can also ultimately lead to alterations in attachment (Melnyk et al, 2001; Swartz, 2005). In sum, due to the transactional relationship between parents and their infants, the negative effects of stress on parents can lead to negative effects for infants as well.

To summarize, parenting in general can be a stressful experience, and parenting a chronically ill infant or one with CHD can be additionally stressful. The stress these parents experience arises from several areas. The inability to orally feed and the subsequent physical growth difficulties these infants have can lead to elevations in parenting stress. The severity of the infant's CHD and the consequential intensive care hospital environment and resultant home care required can contribute to parenting stress as well. The environment includes not only the ICU and hospital encounter but also the socio-demographic situation of the family and the resources parents have available to them. Consequently, when examining parenting stress, it is important to be cognizant of the obvious and obscure contributing factors.

Congenital Heart Defects (Severity of Illness)

Congenital heart defects (CHD) are structural problems that arise from the abnormal formation of the heart or major blood vessels in utero. There are at least fifteen distinct types of congenital defects recognized, with many more anatomic variations (Rosamond et al, 2007). These defects range in severity from a pinhole size ventricular septal defect (VSD) that may spontaneously close to very complex single ventricle lesions such as hypoplastic left heart syndrome (HLHS) requiring multiple surgeries to repair. In 1000 live births, approximately nine of those infants will have a CHD, which comes to about 36,000 infants per year in the United States (US) (Rosamond et al, 2007; Steltzer, Rudd, & Pick, 2005). Present estimates indicate there are one million individuals currently living with CHD (Green, 2004). Congenital heart defects are the most common cause of infant death from birth defects in the US, in that, 30% of infants who die from a birth defect, have a heart defect (Rosamond et al, 2007). One of the many

challenges accompanying infants with CHD is their growth and the many nutritional implications, which are directly related to the severity of their defect (Steltzer et al., 2005). Though defects vary in size and severity, the impact a CHD diagnosis has on a family can be devastating whether it happens prenatally or after the infant is born.

Diagnostic capabilities have provided the ability to detect CHD prenatally. The benefits of prenatal diagnosis are equivocal. The terms, "shock' and "burden" are often used when parents describe their infant's CHD diagnosis. Some parents view the knowledge of a CHD during the remainder of pregnancy as a burden, something they would obsess about but have no control to change. Others welcome the information and see it as a way to plan and prepare for what is to come, and this in some small way gives them a sense of control (Brosig, Whitstone, Frommelt, & Frisbee, 2007). Despite months and months of planning, whether parents find out at twenty weeks gestation or several weeks after delivery, little truly prepares them for the realities of having an infant with a CHD and the resultant increased parental stress they will experience (Skari et al, 2006).

Congenital heart defects are classified into two major groups, acyanotic and cyanotic. Both types of defects present challenges related to the growth and development of these infants. Acyanotic defects are typically associated with increased pulmonary blood flow or obstruction across the heart valves (Steltzer et al., 2005). Lesions of this type that cause left-to-right shunting result in significant volume overload of either the left ventricle, right ventricle, or both depending on the specific abnormality, which inevitably leads to pulmonary overcirculation (Steltzer et al., 2005). The acyanotic lesions most prone to this pulmonary overcirculation, with accompanying risk for growth failure, include VSD, patent ductus arteriosus (PDA), atrial septal defect (ASD), atrio-

ventricular (AV) valve regurgitation, and less commonly semilunar valve regurgitation (Steltzer et al., 2005). The effect of significant shunting leads to height and weight disturbances and the type of defect can dictate the potential for growth failure (Steltzer et al., 2005). Acyanotic patients, particularly those with increased pulmonary blood flow and resulting congestive heart failure (CHF), exhibit more growth delay (Peterson & Wetzel, 2004). Also the weight of children with acyanotic lesions is typically more affected than their height (wasting) in contrast to cyanotic children who often have similar or greater retardation in height than weight (stunting) (Nydegger & Bines, 2006; Peterson & Wetzel, 2004; Steltzer et al., 2005).

Cyanotic defects are associated with right-to-left shunting and result in hypoxemia (Steltzer et al., 2005). These lesions include double outlet right ventricle (DORV), transposition of the great arteries (TGA), tetrology of Fallot (TOF) with and without pulmonary atresia, tricuspid atresia (TA), and HLHS and are associated with disturbances in weight and height (Steltzer et al., 2005). There is a direct relationship between hypoxemia and growth, but the degree of cyanosis has not been found to correlate with the severity of growth impairment; however, the degree of growth impairment is closely related to the severity of the hemodynamic impairment (Steltzer et al., 2005; Varan, Tokel, & Yilmaz, 1999). It is the duration of the hypoxemia in years, not severity that is felt to play a significant factor in growth retardation, and if this hypoxemia is accompanied by CHF, growth (weight and length/height) is even more severely affected (Steltzer et al., 2005).

Any hemodynamic impairment resulting in CHF negatively affects the nutritional status of infants with CHD (Steltzer et al., 2005). CHF often correlates with clinical

findings of tachypnea, hepatomegaly and tachycardia, and it influences growth by affecting caloric intake, increasing metabolic rate, altering gastrointestinal (GI) function and by causing malabsorption (Steltzer et al., 2005). Lesions that commonly result in CHF include: HLHS, TGA, PDA, total anomalous pulmonary venous return (TAPVR), critical valvular aortic stenosis, coarctation of the aorta, and VSD (Steltzer et al., 2005). The larger the left-to-right shunt, the greater the potential for excessive pulmonary blood flow, increased pulmonary artery (PA) pressure, increased blood return to the left heart, and elevation of left ventricular end-diastolic volume and pressure; it is this condition of high output hemodynamics that causes the hypermetabolism that leads to growth failure (Steltzer et al., 2005).

There is much debate as to how the severity of CHD should be classified. The most prominent method, springs from a major multi-institutional effort to measure the complexity of congenital heart surgery, the Risk Adjustment in Congenital Heart Surgery-1 (RACHS-1) system (Jacobs, Wernovsky, & Elliot, 2007). The RACHS-1 is strongly associated with in-hospital mortality and length of stay, and its predictive value is higher than that of other complexity scores (Al-Radi et al, 2007; Kang, Tsang, Elliot, de Leval, & Cole; 2006). Though severity of illness may be a less critical component of successful adaptation than maternal perceptions or the resulting quality of the mother-child relationship (DeMaso et al, 1991), it remains an important variable to capture and consider when examining parenting stress. The diagnosis and classification of CHD, the resultant intensive care, surgeries, at home therapies and all their sequelae, such as growth and developmental delays, all serve as potential elevators of parenting stress.

Infant temperament

Infants enter the world with various temperaments, most notably classified into three categories by Thomas and Chess, "easy", "slow to warm up", and "difficult" (1977). Infants with "difficult" temperaments present a greater challenge for parents and this type of temperament contributes negatively to parenting competence and stress, and is associated with more problem behavior later in life (Secco & Moffatt, 2003; Gutteling 2005). Parental distress significantly increases the likelihood of an infant being perceived as difficult (Mäntymaa, Mirjami et al, 2006; Sheinkopf et al., 2006). The directionality of this relationship has not been fully explicated. It may be that (di)stressed parents perceive their infants as difficult which leads to more stress, which leads them to further perceive their infants as difficult, thus creating a vicious cycle. In fact Sheinkopf et al found infant temperament and parental attitudes have reciprocal effects over time (2006). As a result, when studying parenting stress, it is equally important to examine infant temperament as well.

Nine temperament categories were established by Thomas and Chess while conducting their New York Longitudinal Study (Chess & Thomas, 1989). The categories are activity level, rhythmicity (regularity), approach or withdrawal, adaptability, threshold of responsiveness, intensity of reaction, quality of mood, distractibility, and attention span/persistence (Chess & Thomas, 1989). These nine dimensions cluster into three behavioral styles: easy, difficult, and slow-to-warm up. The "easy child" shows regularity of biological functions, positive approach responses to new stimuli, high adaptability to change, and mild or moderately intense mood expressions, which are mostly positive (Chess & Thomas, 1989). Forty percent of their sample fit into this

category. Ten percent of their sample was temperamentally "difficult". These children show irregularity in biological functions, have negative withdrawal reactions to new stimuli, adapt slowly to change and typically have many negative emotional expressions of loud intensity. The slow-to-warm up child also tends to show negative withdrawal responses to novelty, slow adaptability to change, and many negative mood expressions in comparisons with other children; they however, have mood expressions that are mild to moderate in intensity and have biological functions that may or may not be irregular. These children made up 15% of the sample and are often labeled as shy. These categories only captured 65% of their sample, demonstrating that not all children neatly fit into one of these three temperamental patterns.

The difficult behavior style is the one most commonly associated with behavioral difficulties and disorders (Chess & Thomas, 1989). This behavior style constellation is composed of 5 dimensions characterizing the infant as: arrhythmic, withdrawing, not adaptable, displaying intense moods that are often negative. Caution however, should be used before applying the label of "difficult" to a developing infant because there is a considerable amount of change occurring during the first year of life, especially for premature infants and possibly for other infants hospitalized early in life (Hughes, Shults, McGrath, & Medoff-Cooper, 2002). Perhaps the more important concept is the fit between infant/child behavior style and parental expectations; this model is known as "goodness of fit" (Chess & Thomas, 1989).

Infant temperament is inborn but may also be influenced by environmental factors such as early illness and subsequent hospitalization. Although it is thought that it may be perceptions of temperament that are more affected by early perinatal crises (severity of

illness leading to increased length of hospital stay) rather than actual temperament characteristics (Spungen & Farran, 1986; Langkamp, Kim, & Pascoe; 1998). This was demonstrated when mothers of preterm infants gave general perceptions of their infants as more difficult than they objectively rated them on the Early Infant Temperament Questionnaire (Langkamp, Kim, & Pascoe; 1998). They found perceptions were more highly predictive of later behavioral problems than actual ratings of temperament, with maternal perceptions of difficult temperament in infancy being associated with increased risk for behavioral problems in preschool (Langkamp, Kim, & Pascoe; 1998). Parent perceptions may contribute to the goodness or poorness of fit between parent and child, with a poor fit increasing the risk for child abuse and vulnerable child syndrome (Langkamp, Kim, & Pascoe; 1998).

Not only does the postnatal environment influence temperament, it is thought that the prenatal environment does so as well. Maternal prenatal stress is associated with temperamental and behavioral problems in toddlers (Gutteling et al, 2005). These same authors also found that fear of having a handicapped child was a predictor of higher levels of restless/disruptive temperament (2005). However, it may be the perceptions of the mothers leading them to think their infant is more difficult (because of the stress) when in fact s/he is not (Langkamp, Kim, & Pascoe; 1998). Receiving a prenatal diagnosis of CHD can be an incredibly stressful situation for parents, and this increased prenatal stress may add another factor to the equation leading parents to consider infants with CHD as more temperamentally difficult.

The only published study found examining temperament in infants with CHD was conducted nearly twenty years ago (Marino & Lipshitz, 1991). They discovered a

relationship between temperament and CHD, in that infants and toddlers with CHD were perceived differently than healthy controls (by their parents) on several dimensions of the Revised Infant Temperament Questionnaire (RITQ) and the Toddler Temperament Scale (TTS). The sample included thirty-six infants, ages 4-8 months, and sixty-one toddlers, ages 12-36 months. The infants were perceived as more withdrawn, more intense, and having a lower threshold for stimulation, and the toddlers as less active, less rhythmic, less intense and more negative. Temperament, though related to environmental differences such as cardiac disease, was not correlated with disease severity. The authors suggest the presence of a mediating factor in the relationship between temperament and illness such as parental perception of the child which influences parental report of temperamental differences. The authors give no concrete recommendations for future research though the mention of parental perception of temperament versus actual temperament suggests an area of further exploration.

The manner in which caregivers react to and cope with challenging infant behaviors can be expected to affect infant development (Sheinkopf, 2006). Infants and toddlers with CHD appear to have some challenging elements of their temperaments, and with their parents experiencing increased levels of stress, this may negatively influence their perceptions of their infants, which may in turn increase parenting stress further. Temperament characteristics are an important part of the parent-infant relationship to study because of their influence on emotional and verbal interactions, which in turn influence social and cognitive abilities (Hughes, Shults, McGrath, & Medoff-Cooper, 2002). Both early temperament and parent-infant relationship quality contribute to the

subsequent psychological/behavioral and physiological functioning of the child (Burgess, Marshall, Rubin, & Fox, 2003).

Demographic variables

Socio-economic status (SES) variables such as education, occupation, and income remain closely correlated with infant developmental outcomes in healthy infants and in those with CHD (Wernovsky, 2006). Maternal education has been found to be the single most important factor in predicting children's educational development (Davis-Kean, 2005). It would stand to reason that SES plays a large role in the nutrition available for infants, and would consequently impact physical as well as mental and psychomotor outcomes.

Another important demographic variable to asses is race/ethnicity. Mortality from CHD is higher and has declined more slowly among Blacks than among Whites (Boneva et al., 2001). Though the prevalence of CHD may be lower in Blacks than among Whites, the death rates remain higher for Blacks (Boneva et al, 2001). Forty-three percent of deaths from CHD occur in infants less than a year of age, and for a number of defects (e.g. TGA, ToF, VSD) Blacks die at younger ages than Whites, often approximately half the age of Whites (Boneva et al, 2001). Though race and ethnicity are acknowledged as social constructs and as such have little to no biologic significance, they are important variables to examine as they relate to racism, access to care, and other health disparities related concepts.

It is also important to examine gender, not only because males and females have different growth trajectories, but parent stress may also be elevated in mothers of male infants when compared to mothers of female infants (Scher & Sharabany, 2005). Such is

the case with post-partum depression (Weinberg, Olson, Beeghly, & Tronick, 2006), but it is not known why this occurs, or if it holds true for mothers of medically fragile infants as well. Further differences between the male and female infants reveal male newborns as less responsive to auditory and social stimuli, less able to maintain eye contact; they experience greater difficulty in maintaining affective regulation, smile less, display more irritability, crying, facial grimacing, and lability of emotional states than female infants (Weinberg, Tronick, Cohn, & Olson, 1999). Death rates from CHD are also higher among boys, especially during infancy, which is partially explained by the higher proportion of boys among infants born with serious CHD (e.g. hypoplastic left heart syndrome, transposition of the great arteries, pulmonary atresia, tricuspid atresia, coarctation of the aorta, and aortic stenosis) (Boneva et al., 2001). Also of note, female gender has been reported as a risk factor for post-operative in-hospital mortality (31% - 51% greater) (Chang, Chen, & Klitzner, 2001; Seifert, Howard, Silbert, & Jobes, 2007). It is not known why this occurs or through what mechanism this phenomenon operates, but it is important to include gender in the analysis of infants with CHD for these reasons.

Growth

It is commonly agreed that growth failure in CHD is one of its most common and challenging consequences (Jackson & Poskitt, 1991; Chi-Wen Chen, Chung-Yi Li, & Jou-Kou Wang, 2004; Peterson & Wetzel, 2004; Steltzer et al., 2005; van der Kuip et al., 2003; Varan, Tokel, & Yilmaz, 1999). What is not so commonly agreed upon is how best to operationally define growth failure. Failure to thrive (FTT), malnutrition, undernutrition, growth failure, and growth deficiency are all terms used to define suboptimal growth in this population. FTT is a diagnosis used to describe impaired

physical growth, especially deficient weight gain that is explained as a consequence of deficiency between energy retention and energy requirements (Ward, Lee, Lipper, 2000; Jackson & Poskitt, 1991; Steltzer et al., 2005). FTT is applied to infants with weights below the 5th percentile and who show significant failure to gain weight at age appropriate rates (falling back two or more standard deviations [SD] on standardized norms-for-age and -gender growth charts) in six month or less (Ward, Lee, Lipper, 2000). Malnutrition has been defined as a state of poor nutrition and growth failure (Steltzer et al., 2005), with FTT being used more in developed countries and malnutrition more in developing countries (Ward, Lee, Lipper, 2000). Another relevant term is catch-up growth, which is the velocity of growth following a time period of impaired growth caused by undernutrition (Steltzer et al., 2005). Growth in infants is a direct reflection of their nutritional well-being and is the single most important parameter used in assessing their health status (Lipman et al, 2004).

Immediately after birth, infants experience a weight loss of about 6% of their birth weight (BW), and occasionally this will reach and even exceed 10% (Steltzer et al., 2005). This weight loss is the result of fluid loss and some catabolism. Healthy neonates gain this weight back by 10-14 days of life (Steltzer et al., 2005). As the infants continue to gain weight, it is expected in healthy full-term infants that weight gain will take place at the pace of about 20-30g/d during the first six months of life (Steltzer et al., 2005). Along with weight gain, the infant grows longer as well; incremental gain in crown-heel length should average about 0.66cm/wk during the first six months of life (Steltzer et al., 2005). Infants also display rapid increases in head circumference (HC), and this head

growth, which averages 0.33cm/wk, correlates well with brain growth (Steltzer et al., 2005).

At birth, most infants with CHD have a normal weight for gestational age (GA), but nutritional and growth problems become evident early in life (Nydegger & Bines, 2006; Steltzer et al., 2005). Weight tends to be more affected than height, but if the nutritional deficit is severe enough and lasts long enough, liner growth will be retarded as well (Steltzer et al., 2005; Witzel et al., 2006). In comparison with healthy infants, fluid losses in the neonate with CHF are 10-15% greater because of tachypnea, emesis, diarrhea, and the anti-congestive management with diuretics (Steltzer et al., 2005). There is an "intimate relationship" between energy intake and expenditure and nutritional status and growth in infancy (Nydegger & Bines, 2006), and when any element of this relationship is unbalanced in any way, negative effects often ensue.

Malnourished infants are more prone to both infectious and non-infectious complications of their disease and/or therapy which can often result in a longer hospital length of stay (Kelleher, Laussen, Teixeira-Pinto, & Duggan, 2006; Nydegger & Bines, 2006). Malnutrition can adversely affect the immune system, which can result in postoperative infections, such as pneumonia and delayed wound healing (Steltzer et al., 2005). Along with infection concerns, malnutrition affects both physical as well as cognitive development.

Concerns for poor nutrition relate to long term outcomes such as brain development and oral-motor skill attainment in addition to physical development (Steltzer et al., 2005). If brain growth and function are at risk from poor nutrition, it is speculated that the greatest risk is likely to be in early life when brain cell multiplication

and development are most rapid (Browne et al., 2005). Prolonged periods of malnutrition may inhibit both brain growth and the infant's opportunities to learn from the environment, thus increasing the likelihood of developmental delays (Jackson & Poskitt, 1991). These negative sequelae could persist as infants with FTT may continue to experience deficiencies in motor skills and IQ for 4-10 years after surgical repair (Chi-Wen Chen et al., 2004). Chronic malnutrition and feeding problems also place infants at risk for poor social cognitive functioning that may disrupt normal parent-child interactions (Imms, 2004), revealing the close link between growth (nutrition) and infant development.

Development

An increased incidence of adverse neurodevelopmental and behavioral outcomes exists for infants and children who survive open heart surgery for complex CHD. These deficits, which include cognitive and intellectual impairment, fine and gross motor delays, mental retardation, learning disabilities, executive function deficits, visual-spatial and visual-motor skills deficiencies, speech and language delays, and behavioral difficulties such as inattention and hyperactivity, can appear later in childhood and lead to long term functional impairments (Ballweg, Wernovsky, & Gaynor, 2007; Brown, et al., 2005; Green, 2004, Wernovsky, 2006). As they progress through school, low academic achievement scores, learning disabilities, behavioral problems, and attention deficit and hyperactivity may result in academic failure, poor classroom and social skills, low self-esteem, behavioral disinhibition, and ultimate delinquency (Wernovsky, 2006). As they get older, the need for special services in school is significantly increased compared to the general population. The combination of developmental delay, academic difficulties,

and behavioral abnormalities represent the most common morbidity affecting the quality of life in school age survivors of CHD (Wernovsky, 2006). There are patient specific factors that contribute to developmental outcomes as well as management specific ones.

Approximately one-third of all children with CHD have other abnormalities in addition to their cardiac disease (Wernovsky, 2006). Some patient-specific factors contributing to adverse developmental outcomes include genetic syndromes such as trisomies 13, 18, and 21; William's syndrome; Noonan's syndrome; CHARGE association; VACTERL, and DiGeorge or 22q11.2 microdeletion (Ballweg et al, 2007). These all have an increased incidence of CHD and are associated independently with developmental delays, which potentially confounds research findings with this patient population (Ballweg et al, 2007; Green, 2004). Sub-chromosomal gene abnormalities are being discovered with increasing frequency in this population, and most studies report worse outcomes in children with associated congenital anomalies compared to children with the same lesion and no anomalies (Wernovsky, 2006). Lower birthweight and younger gestational age at time of surgery, and palliative (as opposed to corrective) surgery are also risk factors for and predictors of worse neurodevelopmental outcomes (Ballweg et al, 2007; Green, 2004).

Socioeconomic (SES) status is perhaps the strongest predictor of neurodevelopmental outcomes (Wernovsky, 2006). The relationship between SES and parental intelligence and outcomes in children with CHD has been established (Wernovsky, 2006). Socioeconomic status and parental IQ predict neurocognitive developmental outcomes (IQ at age 5) after cardiopulmonary bypass (CPB) (Ballweg et

al, 2007). Higher SES predicted higher IQ and academic performance for 133 patients who underwent the Fontan procedure (Ballweg et al, 2007).

Another patient-specific factor is central nervous system (CNS) development. There is increasing evidence CNS development in utero is abnormal in children with CHD. The CNS and cardiovascular systems form nearly simultaneously in early gestation; abnormalities in one system increase the likelihood of having problems in the other (Wernovsky, 2006). Infants with CHD have an increased incidence of structural brain abnormalities (periventricular leukomalacia [PVL], microcephaly, incomplete closure of the operculum, and cerebral dysgenesis) that may be caused by abnormal fetal flow patterns, and postnatal cerebral blood flow (CBF) is dramatically reduced in some infants as well (Brown et al, 2005; Ballweg et al, 2007; Wernovsky, 2006). The PVL and microcephaly in some newborns with CHD may be evidence of ischemia related to this low CBF (Green, 2004). The presence of PVL is associated with low baseline CBF and decreased carbon dioxide reactivity, which are associated with poor neurodevelopmental outcomes and a higher risk of death (Ballweg et al, 2007).

Many of the same factors associated with adverse neurodevelopment, including the development of PVL, are associated with hypoxemia and hypotension postoperatively (Ballweg et al, 2007). Oxygen saturations below normal potentially compromise delivery of oxygen to the brain (Wernovsky, 2006). Since 50% of brain growth occurs during the first year of life, prolonged hypoxemia, congestive heart failure, and failure to thrive are likely to affect development (Brown et al., 2005). Though chronic and intermittent hypoxemia are associated with adverse effects on development, behavior, and academic achievement, even in children with structurally normal hearts (chronic lung disease, sleep

disordered breathing, high altitude), it is difficult to measure the effects of hypoxemia in isolation (Wernovsky, 2006).

Newborns and infants with CHD at times have neurodevelopmental abnormalities *before* surgery such as hypotonia, hypertonia, jitteriness, motor asymmetries, and an absent suck. This is important due to the strong association between preoperative and postoperative neurodevelopmental status (Brown at al., 2005; Green, 2004). Injury to the CNS in infants with CHD is characterized by abnormalities of tone, feeding difficulties, delays in major motor milestones, and abnormalities in speech (Wernovsky, 2006). The brain of full term neonates with CHD structurally resembles that of a preterm neonate; consequently, school age survivors of complex heart surgery have developmental findings very similar to survivors of preterm birth, which suggests a similar pathological response to injury (Wernovsky, 2006).

Pre-, intra-, and postoperative management factors also contribute to developmental outcomes. Prenatal diagnosis of the congenital defect enables early initiation of prostaglandins which maintain the patency of the ductus arteriosus, preventing acidosis, which is related to later neurologic injury (Ballweg et al, 2007). Cardiopulmonary bypass (CPB) has been implicated in neurologic injury and speech dysfunction, potentially caused by embolic complications and/or the activation of a variety of inflammatory pathways, all of which can lead to short and long term cognitive defects (Ballweg et al, 2007; Brown et al, 2005; Green, 2004). Deep hypothermic circulatory arrest (DCHA) is not without risk either; it has been associated with cognitive defects as well, particularly with prolonged duration (Brown et al., 2005). While examining the relationship of surgical approach to neurodevelopmental outcomes in

HLHS, no relationship between surgical strategy and any outcome measure of developmental outcome was found (Mahle et al., 2006). They did find deficits prevalent among school-aged children with HLHS regardless of surgical approach, and complications resulting in a prolonged hospitalization at the time of their initial operation were associated with neurodevelopmenal status at school age (2006).

Length of stay (LOS) in the hospital and the intensive care unit may be important markers of late neurologic morbidity. Significant determinants of LOS include pre-operative intubation, longer total support time, postoperative re-intubation, hypotension, arrhythmia, sepsis, and higher inotropic support (Ballweg et al, 2007). Longer postoperative LOS is associated with worse cognitive function (lower full scale IQ scores and verbal performance scores) even when adjusting for factors known to adversely affect long term outcomes (seizures, intraoperative support duration, reoperations and other postoperative events, and sociodemographic variables) (Brown et al, 2005; Mahle, 2006; Wernovsky, 2006). There is also an association between longer LOS and lower Psychomotor Development Index (PDI) on the Bayley Scales of Infant Development at one year of age (Ballweg et al, 2007).

Although the majority of children with CHD have normal neurodevelopmental outcomes, especially those without coexisting CNS abnormalities at birth, the far reaching implications for the children experiencing detrimental outcomes highlights the importance of this area of research (Ballweg et al, 2007). It is important to keep in mind that developmental studies in infants have limited predictive validity for long term outcomes, both in patients with and those without CHD (Wernovsky, 2006), so larger longitudinal studies in this area are needed. The causes of the potential academic

difficulties survivors of CHD face are multifactorial, additive and incompletely understood (Wernovsky, 2006). Along with longitudinal studies, other demographic variables such as gender, race, and ethnicity should be explored because to date they have not been well studied as determinants of neurodevelopmental outcomes (Ballweg et al, 2007).

Gaps

It is well known that stress is increased in parents of infants with CHD, and the factors that impact this stress are myriad. The infant's severity of illness and the resulting growth and development issues can further contribute to parenting stress. Infant temperament, notably infants with "difficult" temperament characteristics can also serve to increase levels of parenting stress. The environment of the hospital and specifically the intensive care unit can elevate stress levels as well. Parenting stress arises not only from infant and hospital environmental factors but also socio-demographic and life factors as well.

The pediatric and neonatal ICU environments have been studied and give some insight into how parents feel and what the most stressful aspects of those environments are, but little has been done to explore the cardiac ICU. The feelings of helplessness parents of infants in the CICU may experience and their implications have also been understudied. Perhaps parents of infants and children in the CICU have similar feelings and experiences as NICU and PICU parents and interventions used in those populations to address these issues can be tailored for the CICU as applicable.

It is known that the severity of CHD contributes to problems with growth and development, which result in increases in parenting stress. If increased levels of

parenting stress have been found to be negatively correlated with development in low birthweight infants, a similar relationship might exist for infants with CHD as well. Also stress as it relates to prenatal CHD diagnosis is another area of potential research. It is quite possible that interventions to reduce parenting stress should be implemented well before the birth of the infant due to the relationship between prenatal stress and "difficult" infant temperament characteristics.

A closer examination of sociodemographic variables such as race/ethnicity and gender is needed as explanations for disparities in outcomes are explicated. It is not known why higher mortality exists for Black infants compared to Whites or why female infants experience higher in-hospital post-operative mortality than males. Further inclusion and exploration of these variables will hopefully yield answers in the near future.

It is known that increasing the caloric intake of infants with CHD helps combat growth failure, though maximizing calories is not always possible due to oral feeding problems. If biopsychosocial relationships of the theoretical framework hold true, there may be additional ways to address growth and development problems in this patient population by modifying parenting stress or other mutable variables. This patient population and phenomenon of interest are ripe with research possibilities. Much has been studied to date, with much yet to be discovered. The role parenting stress may play in physical growth and neurodevelopmental outcomes has not been explored, and if a significant relationship exists, possibilities for intervening also exist.

Chapter 3: METHODS

Introduction

This chapter discusses the parent study upon which the proposed study is built. The study sample and setting are described as well as a detailed description of each of the variables and instruments to be utilized in the proposed study. The data analysis plan is presented, and the chapter concludes with a summary of human subjects' protection.

Parent Study

This study will build upon the parent study "Feeding Behaviors and Energy Cost in Infants with Congenital Heart Disease" (CHD Feeding Study). This parent study focuses primarily on developing a predictive model of failure to thrive in this infant population. The CHD Feeding Study seeks to examine both sides of the energy balance equation, feeding and energy intake as well as energy expenditure to determine the contributions of both in the growth failure or success in infants with CHD.

The parent study examined infants during the first twelve months of life at five time points- newborn (during the first six weeks of life), three, six, nine, and twelve months of age. Each visit measured a differing set of variables and ranged in time from thirty minutes to four or five hours in length (see Table 1). One of the aims of the CHD Feeding Study was to determine which aspects of feeding performance (suck/swallow/breathe coordination, etc.) are most subject to disruption in this patient population. To that end, feeding performance was measured by using a specially designed bottle and nipple system at the newborn, three, and six month visits. The newborn visit also included anthropometrics or body measurements and a measurement of body composition via Total Body Electrical Conductivity (TOBEC).

The three month visit included, along with the feeding, anthropometrics, and TOBEC, measures of energy expenditure while sleeping (Resting Energy Expenditure or REE) and total daily energy expenditure (TEE). The REE was measured by way of respiratory calorimetry and TEE by utilizing the doubly labeled water (DLW) technique. At this visit parents also completed the Parenting Stress Index (PSI) and the Early Infancy Temperament Questionnaire (EITQ). The six month visit included the feeding and anthropometrics along with the Bayley Scales of Infant Development- 2nd Edition (BSID-II) administered by a developmental psychologist. The parents also completed the PSI and the Infant Temperament Questionnaire (ITQ). The nine month visit, the shortest of the series, consisted of only anthropometrics and the PSI. The twelve month visit, the longest of the series contained elements from each of the previous visits: REE and TEE, anthropometrics, TOBEC, BSID-II, PSI, and the Toddler Temperament Scale (TTS). Three-day diet records of food intake were kept after each visit and medications the infants were taking were recorded at each visit as well. Infants with CHD as well as healthy controls were examined. The parent study contains several variables, but this proposed study seeks to only examine a few (in bold italics in Table 1) in order to determine the relationships between parenting stress, growth and development of infants with CHD.

Newborn Visit	3 Month Visit	6 Month Visit	9 Month Visit	12 Month Visit
Feeding Anthropometrics TOBEC Diet record	Feeding ***Anthropometrics*** TOBEC REE TEE ***PSI*** ***Temperament*** Diet record	Feeding ***Anthropometrics*** ***PSI*** ***Temperament*** ***BSID-II*** Diet record	Anthropometrics PSI Diet record	Anthropometrics REE TEE PSI Temperament BSID-II Diet record

Table 1
Variables for proposed study
in ***bold italics***

Sample and Setting

Sample

The infants with CHD were all recruited from the cardiac intensive care unit of the Children's Hospital of Philadelphia (CHOP). To meet subject inclusion criteria for the CHD Feeding Study infants: underwent corrective or palliative surgery for their defect within the first six weeks of life, were ≥36 weeks gestation, ≥2500 grams, without multiple congenital anomalies other than their cardiac lesion, without a documented or suspected genetic syndrome (except DiGeorge and 22q deletion), and without craniofacial or gastrointestinal anomalies that could interfere with feeding, digestion, and growth. Parents who were unable or unwilling to return for follow up at the CHOP were excluded. Control infants were recruited from the CHOP faculty practice, CHOP primary care practice, and word of mouth.

Setting

Subjects were recruited from the Cardiac Intensive Care Unit (CICU) at the Children's Hospital of Philadelphia (CHOP). This unit, one of the largest and busiest in the world, performs more than 1,000 cardiothoracic surgeries (including 500 open heart procedures) per year. They also perform more than 1000 cardiac catheterizations a year. The unit contains more than 50 physicians and surgeons and more than 350 other staff including nurses, respiratory therapists, social workers, and child life therapists (About the Cardiac Center; http://www.chop.edu/consumer/jsp/division/generic.jsp?id=87547).

All outpatient visits took place in the General Clinical Research Center (GCRC) and the Nutrition and Growth Laboratory (NGL) at the CHOP. The GCRC is staffed with registered nurses, phlebotomists, and technicians skilled in data collection for

research projects throughout the entire hospital. When the infants arrived for each outpatient visit, the infant's vital signs were assessed and documented by either a GCRC nurse or a CHD Feeding Study nurse. The patient and family were then escorted down the hall to the NGL where one of the trained growth technicians assessed the required variables for the appropriate time point. Parents were given meal vouchers for lunch in the cafeteria and parking validation if needed. Once the questionnaires and diet records were returned, parents received a gift certificate to a children's toy store of varying amounts depending on the time point ($25-$100).

Variables and Instruments

Severity of illness

Risk Adjustment for Congenital Heart Surgery 1 (RACHS-1)

The increase in outcomes research led to the need to develop a method of risk adjustment due to the varied nature and range of congenital heart defects (Jenkins et al, 2002; Jenkins, 2004). A panel of experts and two large multi-institutional data sets were used to create the Risk Adjustment for Congenital Heart Surgery-1 (RACHS-1), which uses surgical procedures as the primary proxy for risk (Jenkins et al., 2002). Cardiac procedures are clustered into six risk categories, with category one representing the lowest risk of in-hospital mortality and six representing the greatest (Jenkins et al., 2002).

This method has been designed to allow a refined understanding of differences in mortality among patients undergoing congenital heart surgery, as would typically be encountered within a pediatric population, and when more than one procedure is performed simultaneously, the procedure with the greatest risk category is used to classify the procedure (Jenkins, 2004). It can be used to evaluate the independent effect

of patient-level factors such as gender, race, or insurance type on in-hospital mortality by taking into account the diversity of anatomy inherent in pediatric CHD, and reducing the anatomical differences to a 6-item ordinal scale (Jenkins et al., 2002). The first step uses the cardiac surgical procedure as a surrogate for diagnosis and the next step groups the procedures together that have a similar risk for mortality (Appendix 1).

Parenting stress

Parenting Stress Index (PSI)

The PSI was developed by Richard Abidin as a measure of stressful parent-child systems in order to plan an optimal intervention program (Willinger et al., 2005). It is a screening and diagnostic assessment technique designed to yield a measure of the relative magnitude of stress in the parent-child system in order to detect systems at risk for the development of dysfunctional parenting behaviors or behavior problems in the child involved (Abidin, 1995). Abidin posited that the Total Stress a parent experiences would be a function of certain salient parental characteristics, child characteristics, and situational variables that directly related to the role of being a parent (Abidin, 1995).

The PSI has undergone six revisions and Abidin expanded the model in 1992 to hypothesize that parenting behavior and child adjustment are influenced by a number of environmental, sociological, behavioral, and developmental variables (Abidin, 1992). It is comprised of 101 items rated on a 5 point Likert scale (strongly disagree to strongly agree) divided into two domains: Child and Parent. The Child Domain (CD) consists of six subscales (47 items): Adaptability, Acceptability, Demandingness, Mood, Distractibility/Hyperactivity, and Reinforces Parent. The Parent Domain (PD) consists of seven subscales (54 items): Depression, Attachment, Restriction of Role, Sense of

Competence, Social Isolation, Relationship with Spouse, and Parent Health. The CD and PD combine to equal the Total Stress (TS) score. There is an optional Life Stress (LS) scale with nineteen yes/no questions depicting certain life events (divorce, death in family, debt, etc.) that gives an index of the amount of stress the parent is currently experiencing outside the parent-child relationship and that is often beyond their control (Browne & Talmi, 2005; Willinger et al, 2005). There is also a defensive responding scale, derived from the Marlowe-Crowne Scale of Social Desirability to assess the extent to which the respondent approaches the questionnaire with a strong bias to present the most favorable picture of him/herself in order to minimize indications of problems or stress in the parent-child relationship (Uzark & Jones, 2003). Higher scores on the PSI are indicative of higher levels of parenting stress.

A score is generated for the Parent and Child domains (and their constituent subscales), Life Stress, as well as a Total Stress. Although each score may be interpreted independently leading to generation of a hypothesis in relation to an individual score, Abidin believes that the clearest picture emerges when the various scores are considered in relation to each other (1995). The total stress score is of primary importance in guiding professional judgments as to whether intervention might be necessary or appropriate for a given parent-child system. It is designed to provide an indication of the overall level of parenting stress an individual is experiencing (Urzark & Jones, 2003). Parents who earn raw total scores at or above 260 should be offered a referral for professional consultation. When this Total Stress score is ≥260, the Child and Parent Domain scores, along with the Life Stress scale are useful in determining the domain from which the stress is emanating, and the subscales of the domains provide even further

breakdown to assist the professional in identifying specific sources of stress in a given domain (Abidin, 1995). High LS scores tend to intensify the total stress the parent is experiencing; when the TS raw score is in the 250 range and the LS raw score is ≥17, a referral to a professional for assistance should be considered (Abidin, 1995). Scores on the PSI greater than the 85th percentile are considered high, and scores between the 81st-84th percentiles are considered borderline. A "normal" stress score would fall between the16th-80th percentiles and a "low" stress score is less than the 16th percentile.

Normative scores for the PSI were developed using a sample of 2,633 mothers with at least one child ranging in age from one month to 12 years and 200 fathers of children ranging from 6 months to 6 years of age. The majority (61%) of the normative sample was recruited from well child pediatric clinics and public school daycare centers in Virginia. Seventy-six percent of the mothers were white, 11% African-American, 10% Hispanic, and 2% Asian, and 95% of the fathers were white and 5% African-American (Abidin, 1995). Twenty-seven percent of the mothers had completed college or graduate school and 23% had some vocational training, whereas 48% of the fathers were college graduates.

Alpha reliability coefficients for the thirteen subscales range from .70 to .84, for the two domains (CD and PD), .90 to .93, and .95 for the TS score. Test-retest reliability over 1 to 3 months was shown to be .63 and .91 for Child and Parent Domains respectively and .96 for the TS score (Thomas, Renaud, DePaul, 2004). It takes less than thirty minutes for the parent to complete the PSI questionnaire.

Infant Temperament

Early Infancy Temperament Questionnaire (EITQ)

The EITQ (Medoff-Cooper, Carey, McDevitt; 1993) is a 76-item parent questionnaire for assessing the nine New York Longitudinal Study temperament characteristics (activity, rhythmicity, approach, adaptability, intensity, mood, persistency, distractibility, and threshold) in one to four-month old infants. The Activity level is the motor component in a given child's functioning, and the amount of movement during bathing, eating, playing, dressing and handling, as well as information concerning the sleep-wake cycle, reaching, crawling, and walking are used in scoring this category. High scores indicate a highly active infant and lower scores indicate inactivity. Rhythmicity or regularity is the predictability and/or unpredictability in time of any function, e.g. sleep-wake cycle, hunger, feeding pattern and elimination schedule. Higher scores indicate an arrhythmic infant and lower scores indicate an infant who is more regular in these functions. Approach or withdrawal is the nature of the initial response to new stimulus, whether it be a new food, new toy, or new person. Approach responses are positive and can be displayed by mood expression (smiling, verbalizations, etc.) or motor activity (swallowing a new food, reaching for a new toy, active play, etc.). Withdrawal reactions are negative and may also be displayed by mood expression (crying, fussing, grimacing, verbalizations, etc.) or motor activity (spitting new food out, pushing new toy away, or moving away from a new person, etc.). Higher scores indicate a withdrawing infant and lower scores an approaching infant. Adaptability is the response to new or altered situations; this is not necessarily the concern with the nature of the initial response, but with the ease with which they are modified in a desired way. Higher scores indicate an infant who is slow to adapt and lower scores indicate an infant who is quick to

adapt. Intensity of reaction is the energy level of response irrespective of its quality or direction. Higher scores indicate an infant who is more intense and lower scores indicate a mildly intense infant. The quality of Mood is the amount of pleasant, joyful, and friendly behavior contrasted with unpleasant, crying, and unfriendly behavior. Higher scores indicate an infant who is more negative in mood and lower scores indicate an infant who is more positive in mood. Attention span and Persistency are two related categories. Attention span concerns the length of time an activity is pursued by the infant, and persistency refers to the continuation of an activity in the face of obstacles in order to maintain the activity. Higher persistence scores indicate an infant with low persistence and lower scores indicate an infant with higher persistence. Distractibility is the effectiveness of extraneous environmental stimuli in interfering with or in altering the direction of ongoing behavior. Higher scores indicate low distractibility or an infant who is difficult to soothe, and lower scores indicate a highly distractible or easily soothed infant. Threshold of responsiveness is the intensity level of stimulation necessary to evoke a discernible response to sensory stimuli, environmental objects and social contacts. Higher scores indicate an infant has a low threshold or is sensitive to stimuli and lower scores indicate a low reactive infant who has a higher threshold for stimuli (Thomas & Chess, 1977).

The majority of the items were adapted from the Revised Infant Temperament Questionnaire to be developmentally appropriate for the very young infant. However, the Persistence/Attention span was difficult to measure in such young infants, so items from the Neonatal Behavioral Assessment Scale provided a framework for developing age-appropriate items for this dimension (Medoff-Cooper, Carey, McDevitt; 1993). Each

item is rated on a six-point scale of frequency of occurrence (almost never-almost always). The standardization population consisted of 404 infants from one pediatric practice. This group consisted of mostly whites (80%) and mothers, with a mean education level of 12.34 ± 3.35 years (range 4-20 years). Means for the nine categories were calculated separately for infants from 1-2 months and 3-4 months of age. Internal consistency for the nine categories ranged from .42 to .76. Test-retest scores, completed between 2 to 3 weeks after the first rating, ranged from .43 to .87, with generally increasing retest levels in the older age group. None of the categories showed significant differences between male and female infants.

This questionnaire is administered during the three month visit. Parents are encouraged to complete the questionnaire (approximate time: 20 minutes) at the visit, but when time did not permit, parents were allowed to take the questionnaire home to complete and return in a pre-paid envelope. The majority of the time the questionnaires were completed by the mother, but whichever parent completed the questionnaires at the first visit was required to complete them for the duration of the infant's enrollment in the CHD Feeding Study. Only mothers' reports were used in the current study.

Revised Infant Temperament Questionnaire (RITQ)

The R-ITQ (Carey & McDevitt, 1978) was used to assess temperament at the 6 month time point. It contains 95 items that measure the nine characteristics of temperament discussed above. The questionnaire assesses several areas of behavior including sleep, feeding, soiling, wetting, diapering, dressing, bathing and responses to new environments. It yields scores for each of the nine characteristics of temperament and five diagnostic cluster groups: easy, intermediate low, intermediate high, difficult,

and slow to warm up. The instrument was standardized on 203 full term infants 4 to 8 months of age. Internal consistency ranged from .49 for distractibility to .71 for approach, with a median of .57 and an internal consistency of .83 for the entire instrument. Internal reliability was satisfactory, with a value of .85 for the entire instrument. The R-ITQ takes less than thirty minutes to complete.

Growth

Anthropometric Assessment.

Body size was assessed as one of the primary indicators of growth and nutritional status. Although the CHD Feeding Study assessed many more anthropometrics, the anthropometric assessment for the current study consisted of weight (accuracy to 0.01 kg), measured on a Scaletronix *(*Scaletronix, White Plains, NY) digital infant scale; length (accuracy to 0.1 cm), measured on an infant length board (Holtain, Crymych, UK); and head circumference measured with a non-stretchable fiberglass tape (accuracy to 0.1 cm) (McCoy, Maryland Heights, MO). Measurement techniques followed the methods described in Lohman et al (1988). All measurements were taken at each time point and recorded in triplicate, with the mean used in analyses. All measurements were obtained by two trained technicians from the Nutrition and Growth laboratory.

Development

Bayley Scales of Infant Development-2nd Edition (BSID-II)

The BSID-II (1993) are composed of 3 distinct scales which measure mental acuity and abilities (Mental Scale), degree of control of body coordination and fine motor skills (Motor Scale), and the child's social and objective orientation to the environment (Behavior Rating Scale). The Bayley scales have been used since 1958 and remain one

of the most accurate and most widely used methods to measure the development of infants and toddlers (Chandlee, Heathfield, Damokosh, Radcliffe, 2002). In addition to re-standardizing the norms, in 1993 the age range of the BSID-II was extended down to one month of age and up to 42 months (Black & Matula, 2000). The items on the BSID-II are arranged in ordinal sequence of increasing difficulty. Raw scores are converted to standardized scored (mean = 100, standard deviation = 15) through tables, yielding a Mental Development Index (MDI) score from the Mental Scale and a Psychomotor Developmental Index (PDI) from the Motor Scale, and the Behavior Rating Scale (BRS) provides information on the child's behaviors during the assessment (Black & Matula, 2000). The MDI assesses the child's language development and problems solving (cognitive) skills and the PDI assess the child's gross and fine motor development (Grigorenko & Sternberg, 1999).

The BSID-II was standardized on 1,700 infants aged 1 to 42 months. One hundred infants were in each of seventeen age groups (50 males and 50 females in each). The sample was stratified according to the 1988 update of the US census by race/ethnicity, parent education, and geographic region. To be included in the normative sample infants had to be full term (36 to 42 weeks gestation) with a birth weight appropriate for gestational age, have no significant medical complications or disabilities, and not be receiving treatment or intervention for disabilities (Black & Matula, 2000). Test-retest reliabilities for time periods of 1 to 16 days range from .83 to .91 for the MDI and from .77 to .79 for the PDI. Stability for the BRS varies greatly depending on the age of the child, ranging from .55 to .90. Inter-rater reliabilities were reported to be .96 for the MDI, .75 for the PDI, and .70 for the BRS. The total test internal-consistency

reliability coefficients are adequate, ranging from .89 (at ages 2.5y and 3y) and .90 (at 3.5y of age). When compared to other measures of general cognitive ability, the concurrent validity of the MDI typically falls in the .70 range, and the highest correlation between the PDI and other indicators of cognitive ability was .59 (Grigorenko & Sternberg, 1999).

For the CHD Feeding study, the scales were administered and scored by doctoral level developmental psychologists at the six and twelve month outpatient visits to the Children's Hospital of Philadelphia with either one or both parents present. Only the MDI and PDI will be used for the proposed study.

Demographics

Demographic characteristics of the infant as well as the parents will be examined. Infant characteristics include race/ethnicity, gender, birth weight, feeding mode at each time point, and a severity of illness indicator (RACHS-1). Parental characteristics include- maternal education and whether of not the infant's heart defect was prenatally diagnosed. Parental characteristics were collected on approximately half of the CHD Feeding Study participants. The majority of mothers in the current study had complete demographic data.

Power Analysis

A post-hoc power analysis was calculated for each regression model to determine the various correlation and r^2 levels detectable with the sample size available with 80% power and an alpha coefficient of .05. This was done using online software statistics calculator verified by a statistician familiar with the parent study.

Data Analysis Plan

Specific Aim 1. Describe the parenting stress, infant temperament, infant growth and development for infants with CHD at three and six months of age

The first specific aim sought to describe parenting stress, infant temperament, growth and development at three and six months of age for infants with CHD. Descriptive estimates of all measures were generated, including measures of central tendency (means, medians), measures of variation (standard deviations, interquartile ranges, ranges), and derived moments of skewness and kurtosis. An analysis of distributional properties was performed to determine if variance stabilizing or normalizing transformations should be applied. Outliers were assessed via visual inspection of distributions and checked for accuracy. Bi-variate correlation matrices were used to estimate the correlation among pair-wise variables assessed in this study. Descriptive statistics of these variables were also generated for control infants and compared to infants with CHD using t-tests, One-way ANOVA, and Chi-square when appropriate.

Specific Aim 2. Examine factors of parenting stress which are associated with and predict growth and development for infants with CHD

The second aim estimated the effect of parenting stress on infant growth and development. Analyses sought to control for parent (maternal education, prenatal diagnosis [PND]) and infant (birthweight [BW], gestational age [GA], length of stay

[LOS], post-operative physiology [POP], race, ethnicity, RACHS-1, feeding mode [FM]) demographic variables and infant temperament variables.

The primary independent variable, parenting stress, was measured by the Parenting Stress Index (PSI). All 17 of the PSI subscale measures were used. They were distractability/hyperactivity, adaptability, reinforces parent, demandingness, mood, acceptability, Child Domain (CD), competence, isolation, attachment, health, role restriction, depression, spouse, Parent Domain (PD), Total Stress (TS), and Life Stress (LS). Each of the PSI measures was assessed when the child was roughly three and six months of age. The time point each variable was assessed is notated by a 3 or 6 after the variable name. Three of the PSI subscales share the same name as three temperament subscales, so the temperament subscales are denoted with "mean" after each variable name (e.g. moodmean3) because the mean scores were used (vs. the z-scores).

The dependant, or outcome, variables were continuous growth and development measures. Growth was captured via the weight, length, and head circumference of the infants at three and six months. Z-scores for each of the three growth measures were calculated using the World Health Organization's growth standards. Refer to Chapter 4 for descriptive statistics of growth measures. Development was measured using Mental Development Index (MDI) and the Psychomotor Development Index (PDI) of the Bayley Scales of Infant-II (BSID-II), which was assessed at six months of age. Descriptive statistics of development measures can be found in Chapter 4 as well. Because this aim sought to examine the effect of stress on growth and development, in addition to cross-sectional effects, temporal effects were assessed by regressing growth and development measures at six months on PSI measures observed at three months.

A.) 3 month Parenting Stress → 3 month Growth (weight, length, HC)

B.) 6 month Parenting Stress→ 6 month Growth (weight, length, HC)

C.) 6 month Parenting Stress → 6 month Development (MDI, PDI)

D.) 3 month Parenting Stress→ 6 month Growth (weight, length, HC)

E.) 3 month Parenting Stress→ 6 month Development (MDI, PDI)

To determine which of the independent variables (IDV) should be used in the regression models outlined above (A-E), a multi-step process was completed. First all of the continuous variables were correlated with each of the dependant variables using bivariate correlations. The independent variables that were correlated with the dependant variable with a significance level of $p< 0.2$ were kept for further analysis (see Appendix 2 for correlation matrices). The IDV that were "kept" were then correlated with each other using a bi-variate correlation to test for multicolinearity with each other. Highly correlated variables were defined as variables correlated with an $R \geq 0.7$. In order to determine which of the highly correlated variables to discard and which to keep for use in the regression model, it was determined that if one of the highly correlated variables was highly correlated with more than one variable it should be discarded. If it was only correlated with one other variable, the independent variable that was more highly correlated with the dependent variable was the one selected for use in the regression model. A similar process was used for the categorical variables. They were correlated with the dependent variable using an independent sample t-test, and IDV with $p< 0.2$ correlations were kept. Multicolinearity was assessed between these IDV using the Chi-square statistic. Variables with a Chi-square statistic significant at the $p\leq 0.01$ were

excluded from the pool of IDV used for regression modeling. The relationship between the continuous and the categorical IDV was examined using independent samples t-tests. Multicolinearity was assumed when variables reached significance levels of $p \leq 0.01$. The same logic for choosing which variable to exclude was used here as well. The remaining variables were used for each stepwise linear regression model presented (see Appendix 2 for complete list of models tested).

Specific Aim 3. Examine factors of growth and development which are associated with and predict parenting stress in infants with CHD

The third aim was to estimate the effect of growth and development on parenting stress. As in the previous aim, analyses controlled for parent and infant demographic variables and infant temperament. The primary independent variables were growth and development, and the dependant variable was parenting stress as measured by the Parenting Stress Index (PSI). Because this aim sought to examine the effects of growth and development on stress, in addition to cross-sectional effects, temporal effects were incorporated by regressing stress measures at six months on growth measures observed at three months. Given that the development variables were collected only at six months, the primary analyses only involved growth variables. Separate general linear models were generated for each PSI outcome measure using the variable selection process outlined in Specific Aim 2.

Specific Aim 4. Identify the changes in parenting stress over the course of the first six months of life

The objective of the final aim of the proposed study was exploratory in nature and sought to identify changes in parenting stress over the first six months of life in infants with CHD and compare to controls. In order to evaluate change in parenting stress over time, differences in three month and six month scores were computed and evaluated using a one-sample-test. Assumptions will be assessed as described above.

Strengths and limitations

The proposed study was the first to examine the psychosocial effects of parenting stress on the biological variable of growth as well as development in infants with CHD. The patient population from which these data will be gathered is fairly homogeneous, which can be a strength and limitation. Due to the demographic similarities of the infants, the outcome variables can be said to truly be effected by the independent variables in question, but this also limits the generalizability of the finding of this study.

Limitations are inherent in the secondary analysis of data. The analysis is confined to the instruments used and variables collected in the parent study, but because the parent study is currently active, the age of these data are not an issue as is the case oftentimes with secondary analysis. Some of the questionnaires were completed at the infant's outpatient visit, while others were completed at home. This could potentially influence the social desirability on the part of the parents completing the questionnaires in the presence of research nurses and assistants. A notation is made regarding where the forms are completed, so this can be factored into the analysis.

Human subjects

Risks and benefits

There are no additional risks or benefits the current study poses above what parents might have incurred from the CHD Feeding Study. Their child's participation in this study may help health care professionals learn more about various factors that influence growth and development beyond energy consumed and expended. The main risks involved with a secondary analysis include issues of privacy (addressed below).

Privacy and confidentiality

When consented, all infants are given an identification number which is used on all data collection forms. Information containing identifiable health information is either contained in a locked file cabinet or in a password protected database. Published data from this study will not include any identifiable information, unless parents have consented to allow their child's photograph to be used. The primary investigator for the current study completed HIPPA training in patient oriented research for the University of Pennsylvania.

Inclusion of women, minorities, and children

Due to the nature of this study, only infants are eligible and enrolled in this study. Though it is not fully understood why, CHD affects females and minorities in lesser rates than white males (Boneva et al, 2001; Benavidez, Gauvreau, Jenkins, 2006), and for this reason it is expected that this gender and racial group will comprise the majority of the sample. However, efforts were made to recruit racial and ethnic minorities as well as female infants when eligible.

The current study was approved by the Institutional Review Board of the University of Pennsylvania.

Chapter 4: RESULTS

Introduction

The purpose of this study was to explore the parenting stress as experienced by parents of infants with CHD during the first six months of life and the relationship between this stress and the growth and development of these infants. The study population, including dyads of mothers and their infants with CHD, is described. Results that address each specific aim are presented, and additional analyses performed to address questions that arose during initial analysis are also presented.

Characteristics of Study Population

In addition to the parent study inclusion criteria, to be included in this study, mother-infant dyads needed to have complete or nearly complete data for the outcome variables: growth, development, and parenting stress. Sixty-one infants met the inclusion criteria, and one control infant was excluded due to a measurement error. Of the 60 infants analyzed, there were 35 infants with CHD and 25 control infants. Of the infants with CHD, 11 had single ventricle physiology and 24 had biventricular physiology.

Maternal Characteristics

The maternal characteristics examined in this study were maternal education and the prenatal diagnosis of the CHD. For all infants with CHD (single and biventricular), 68% of their mothers had at least a college degree. Seventeen percent of those had obtained post-graduate degrees. Fifty-four percent of the mothers of infants with single ventricle (SV) physiology had obtained college degrees or greater. For infants with biventricular (BV) physiology, 75% of their mothers had obtained at least a college degree. Seventy-six percent of the mothers of control infants had obtained at least a

college degree. There were no statistically significant differences between the groups (Table 1).

The other maternal characteristic measured was the prenatal diagnosis of the infant's CHD. For all infants with CHD, 49% were diagnosed prenatally. When prenatal diagnosis was examined by physiology, it was found that 82% of infants with SV physiology were diagnosed prenatally compared to 33% of infants with BV physiology. When these two groups were examined using independent samples T-tests, this difference was significant (p=.046) (Table 1).

Infant Characteristics

The infant characteristics examined were: age at time of visit, birthweight, gestational age, length of hospital stay, gender, race, ethnicity, severity of illness via the RACHS-1, feeding mode at 3 and 6 months, and infant temperament at 3 and 6 months. For all infants with CHD, the mean birthweight was 3415g ±515, 3385g ±483 for infants with SV physiology, 3428.5g ±538 for infants with BV physiology, and 3490g ±673 for control infants. The differences between the birthweight were not significant. The average gestational age (GA) for all infants with CHD, infants with SV and BV physiology and control infants was 39 weeks (Table 2). For all infants with CHD, 69% were male. Of the infants with CHD, 82% of the infants with SV physiology were male and 62.5% of infants with BV were male. This was a statistically significant difference (p=.011). Sixty-four percent of the control infants were male (Table 2).

Regarding race and ethnicity, 97% percent of all CHD infants were White and 9% were Hispanic, and 3% of the infants were Black. Ninety-one percent of infants with SV physiology were White, 9% were Hispanic, and 9% of these infants were Black. All the

infants with BV physiology were White and 8% of them were Hispanic. More than a third of these infants were of "unknown" ethnicity. Seventy-six percent of the control infants were White, 4% were Hispanic, and 24% of these infants were Black (Table 2).

The severity of illness for the infants with CHD was measured using the RACHS-1. This variable was dichotomized into ≤ 3 and > 3 for the purposes of analysis based on acuity level of procedure (see Appendix 1 for list of procedures), with higher numbers indicating greater severity of illness. Others have used the same cut point to dichotomize the RACHS- 1 (Polito et al, 2008). For all infants with CHD, 43% of them had RACHS-1 scores > 3. For the infants with SV physiology, 82% had scores > 3. For infants with BV physiology, 25% of them had scores > 3. This was a statistically significant difference (p=.000) (Table2). Related to severity of illness was the infant's length of hospital stay. For all infants with CHD, their median length of stay (LOS) was 14 days ±13. For infants with SV physiology, their median LOS was 22 days ±11, and for infants with BV physiology, their median LOS was 13 days ±14. The LOS was significantly different between the two groups (p=0.02) (Table 2).

The mode of feeding was examined at 3 and 6 months of age for all infants. For all CHD infants, 89% were feeding by mouth (PO) at 3 and 6 months of age. For infants with SV physiology, 82% of them were fed PO at 3 and 6 months. For infants with BV physiology, 92% of them were fed PO at 3 and 6 months. These variables were dichotomized, PO and non-PO in order to test for significant differences between the groups, and none was found (Table 2).

Infant temperament was also measured at three and six months of age. Descriptive statistics for each of the nine temperament categories are presented in Tables 3 and 4 with

notations of scores that were significantly different between groups denoted with an asterisk. The two subscales that were significantly different between subjects and controls at 3 months were Intensity and Distractibility (Table 3). Infants with SV physiology and infants with BV physiology were less intense than control infants (p=.026 and p=.008 respectively). For the Distractibility subscale, parents of infants with SV physiology had significantly higher distractibility scores (p=.028) than control infants. This means the parents of infants with SV physiology find them more difficult to soothe than parents of control infants. There were no significant differences between infants with SV and BV physiology at 3 months and no significant differences between any of the three groups at 6 months (Table 4).

Diagnostic criteria for the "difficult" behavioral style require a score higher than the test mean in 4-5 of the 5 temperament subscales that comprise the difficult child constellation: rhythmicity, approach, adaptability, intensity, and mood. Two of these subscale scores, including intensity must also be >1 SD above the mean. At 3 months one infant from each group met the criteria (Table 3a). This was 9% of the sample of infants with SV physiology and 4% of the other two groups. At 6 months the number of infants who met these criteria increased. Two infants with SV physiology (18%), 2 with BV physiology (8%), and 3 control infants (12%) (Table 4a).

Table 1. *Maternal characteristics*

Characteristic	**Subjects (N=35) mean [SD]**			**Controls {N=see below} mean [SD]**
	All Infants with CHD	Single Ventricle N= 11	Bi-ventricular N= 24	
Maternal education				
HS graduate	9% (3)	9% (1)	8% (2)	8% (2)
Partial college/trade school	9% (3)	18% (2)	4% (1)	16% (4)
College graduate	51% (18)	36% (4)	58% (14)	36% (9)
Post-graduate degree	17% (6)	18% (2)	17% (4)	40% (10)
Prenatal Diagnosis				
Yes	49% (17)	82% (9)*	33% (8)*	N/A

Independent samples t-test used for significance between SV & BV, * $p<0.05$ ** $p<0.01$
Percent totals may not equal 100 due to rounding

Table 2. *Infant characteristics*

Characteristic	**Subjects (N=35) mean [SD]**			**Controls {N=see below} mean [SD]**
	All Infants with CHD	Single Ventricle N= 11	Bi-ventricular N= 24	
Age at visit (months)				
Three month	3.06 [.364]	3.01 [.42]	3.09 [.34]	3.13 [.413]
Six month	6.32 [.57]	6.44 [.73]	6.27 [.49]	6.24 [.416] N=25
Birthweight (g)	3415 [515]	3385 [483]	3428.5 [538]	3490 [673] N=22
Gestational age (weeks)	39.1 [1.04]	39.1 [.94]	39.1 [1.1]	38.9 [1.6] N=19
Length of stay (days) Median	14 [13]	22 [11]*	13 [14]	N/A

Control Ns differ due to missing data
Mann Whitney U used for significance, * $p<0.05$

Table 2. *Infant characteristics continued*

Characteristic	**Subjects (N=35)**			**Controls (N=25)**
	All Infants with CHD	Single Ventricle N= 11	Bi-ventricular N= 24	
Gender				
Male	69% (24)	82% (9)**	63%(15)**	64% (16)
Race				
African-American	3% (1)	9% (1)	0%	24% (6)
White	97% (34)	91 (10)	100% (24)	76% (19)
Ethnicity				
Hispanic	9% (3)	9% (1)	8% (2)	4% (1)
Non-Hispanic	63% (22)	82 (9)	54% (13)	92% (23)
Unknown	29% (10)	9% (1)	38% (9)	4% (1)
RACHS-1				
≤ 3 (2 or 3)	57% (20)	18% (2)**	75% (18)**	N/A
>3 (4, 5, or 6)	43% (15)	82% (9)	25% (6)	
Feeding mode				
Three months				
PO	89% (31)	82% (9)	92% (22)	N/A
NG	6% (2)	9% (1)	4% (1)	
GT/JT	3% (1)	--	4% (1)	
NG/PO	3% (1)	9% (1)	--	
Six months				
PO	89% (31)	82% (9)	92% (22)	
NG	--	--	--	
GT/JT	9% (3)	9% (1)	8% (2)	
NG/PO	3% (1)	9% (1)	--	

Independent samples t-test used for significance between SV & BV, * $p<0.05$ **$p<0.01$
Percent totals may not equal 100 due to rounding

Table 3. *Descriptive statistics for 3 month Infant Temperament (EITQ)*

Temperament characteristic	**Subjects N=35 Mean [SD] z-Scores**			**Controls N=25**
	All Infants with CHD	*Single Ventricle N= 11*	*Biventricular N=24*	
Activity	3.75 [.90] -.05	3.69 [.67] -.14	3.78 [1.0] -.01	3.98 [.73] .26
Rhythmicity	3.09 [.81] .23	3.15 [.94] .22	3.06 [.77] .23	2.88 [.73] -.03
Approach	2.69 [.75] -.19	2.69 [.75] -.19	2.69 [.76] -.19	2.59 [.60] -.32
Adaptability	2.47 [.72] .12	2.61 [.76] .35	2.4 [.72] .02	2.53 [.59] .22
Intensity	2.81 [.87] -.07	2.76 [.87]* A -.04	2.84 [.89]** B -.09	3.60 [.84] -.53
Mood	2.89 [.80] .17	3.14 [.78] .56	2.78 [.80] -.02	2.65 [.52] -.23
Persistence	2.09 [.55] -.74	2.10 [.66] -.72	2.09 [.50] -.75	1.95 [.51] -.99
Distractibility	2.19 [.72] -.37	2.51 [.89]* A .17 *A	2.05 [.59] -.62	1.92 [.50] -.85
Threshold	4.26 [.70] -.11	4.08 [.54] -.43	4.34 [.75] .03	4.1 [.88] -.39

One-way ANOVA used for significance, * p<0.05 **p<0.01
A= difference between SV & control
B= difference between BV & control

Table 3a. *Infants who met "difficult" child criteria at 3 months*

All infants with CHD	Infants with SV physiology	Infants with BV physiology	Control infants
2 (6%)	1 (9%) male	1 (4%) female	1 (4%) female

Table 4. *Descriptive statistics for 6 month Infant Temperament (ITQ)*

Temperament characteristic	**Subjects N=35 Mean [SD] z-Scores**			**Controls N=25**
	All Infants with CHD	*Single Ventricle N=11*	*Biventricular N=24*	
Activity	4.12 [.64] -.51	4.15 [.52] -.44	4.1 [.70] -.54	4.34 [.53] -.10
Rhythmicity	2.61 [.59] .37	2.78 [.49] .62	2.53 [.62] .26	2.64 [.62] .41
Approach	2.56 [.67] .39	2.56 [.71] .38	2.55 [.66] .39	2.50 [.63] .29
Adaptability	2.24 [.52] .38	2.06 [.59] .07	2.33 [.47] .52	2.26 [.57] .41
Intensity	3.51 [.71] .13	3.63 [.53] .29	3.46 [.78] .05	3.46 [.73] .06
Mood	2.91 [.65] .16	3.2 [.41] .58	2.78 [.70] -.04	2.88 [.56] .11
Persistence	3.02 [.68] -.01	2.87 [.61] -.19	3.09 [.72] .08	3.22 [.98] .24
Distractibility	2.4 [.50] .27	2.29 [.51] .09	2.44 [.50] .35	2.29 [.57] .11
Threshold	3.89 [.49] .14	3.82 [.45] .04	3.93 [.52] .18	3.60 [.78] -.24

Table 4a. *Infants who met "difficult" child criteria at 6 months*

All infants with CHD	Infants with SV physiology	Infants with BV physiology	Control infants
4 (11%)	2 (18%) Both males	2 (8%) 1 male, 1 female	3 (12%) 2 males, 1 female

Data Analysis

Specific Aim 1

The objective of the first specific aim was to describe parenting stress, infant temperament, infant growth and development at 3 and 6 months of life. Parenting stress was examined at 3 and 6 months of age. Infant temperament data were used as one of the infant characteristics used as a control variable (discussed above). Growth was measured at 3 and 6 months of age, and development was measured at the 6 month time point.

Parenting Stress. Table 5 displays the descriptive statistics for the each of the 17 subscales of the Parenting Stress Index (PSI). Significant differences between the 3 groups (SV, BV, and control) are noted by an asterisk. At 3 months of age, there were significant differences between subjects and controls in three subscales - Demandingness, Competence, and Attachment. In general, mothers of infants with SV physiology experienced higher stress due to their child's demandingness, their perceived competence as a parent and their attachment with their infant. Specifically, parents of infants with SV physiology had Demandingness subscale scores that were on average 5.16 points higher than parents of control infants (p=.009), and parents of infants with BV physiology had scores 3.37 points higher than parents of control infants (p=.036). For the Competence subscale, parents of infants with SV physiology had scores 6.43 points higher than parents of infants with BV physiology (p=.003 ANOVA and p=.026 t-test) and 5.1 points higher than parents of control infants (p=.019). For the Attachment subscale, parents of infants with SV physiology had scores 2 points higher than infants with BV physiology (p=.001) and 2.33 points higher than parents of control infants (p=.046).

At the 6 month time point there were three PSI subscales with significant differences between scores for infants with CHD: Acceptability, Competence and Life Stress. In general, mothers of infants with SV physiology experienced more stress regarding the acceptability of their infant, regarding their competence as a parent and more life stress. Specifically, for the Acceptability subscale, parents of infants with SV physiology scored 2.99 points higher than parents of control infants (p=.022). On the Competence subscale, parents of infants with SV physiology scored 3.23 points higher than parents of infants with BV physiology (p=.027). In terms of Life Stress, parents of infants with SV physiology scored 5.86 points higher than parents of infants with BV physiology (.044) and 6.34 points higher than parents of control infants (p=.026) on this subscale. The means, ranges and standard deviations for all the subscales can be found in Table 5.

Scores on the PSI greater than the 85th percentile are considered high. Six of the subscales have a "borderline" classification, the 81st-84th percentiles (Adaptability, Child Domain, Depression, Parent Domain, Total Stress, and Life Stress). A "normal" stress score would fall between 16th-80th percentiles and a "low" stress score is less than the 16th percentile. The majority of parents in each of the three groups scored in the "normal" range (Table 6). However, parents of subjects (infants with SV and BV physiology) had significantly more "high" stress scores on the Demandingness subscale at 3 months than parents of control infants (p=.045). Parents of infants with SV physiology also had significantly more "high" stress scores on the Attachment subscale at 3 months than infants with BV physiology and control infants (p=.005). Parents of infants with SV

physiology also experienced higher Life Stress at 6 months than infants with BV physiology and controls (p=.014).

Table 5. *Descriptive statistics for Parenting Stress Index*

Scale/Subscale	**Subjects Mean [SD]**						**Controls Mean[SD]**
	N	All infants with CHD	**N**	Single Ventricle	**N**	Bi-ventricular	**N**	
Three Months								
Child Domain	26	97.6 [19.3]	10	102.8 [22.6]	20	95 [17.5]	25	88.5 [12.8]
Distractibility/ Hyperactivity	32	23.4 [3.4]	10	25.5 [3.6]	22	22.5 [2.9]	25	23.4 [4.2]
Adaptability	33	25.3 [5.3]	10	26 [5.6]	23	25 [5.2]	25	24.3 [3.9]
Reinforces parent	33	8.42 [3.4]	10	8.9 [5.0]	23	8.2 [2.6]	25	6.72 [.94]
Demandingness	28	19.4 [5.2]	10	20.6 [4.9]**A	21	18.8 [5.3]*B	25	15.4 [3.4]
Mood	35	9.43 [3.0]	11	10.2 [3.3]	24	9.1 [2.8]	25	8.32 [2.0]
Acceptability	33	11.1 [3.1]	10	11.4 [3.8]	23	11 [2.9]	25	10.3 [2.7]
Parent Domain	34	111.8 [20.7]	10	119.4 [23.9]	24	108.7 [18.9]	24	110.8 [18.2]
Competence	34	23.8 [6.0]	10	28.3 [7.2]*A, **C	24	21.9 [4.3]	25	23.2 [4.2]
Isolation	35	12.3 [3.3]	11	13.2 [3.4]	24	11.9 [3.2]	24	12.0 [4.0]
Attachment	35	11.6 [2.8]	11	13 [3.9]*A, **C	24	11 [1.9]	24	10.7 [2.5]
Health	35	12.4 [2.7]	11	12.4 [3.0]	24	12.4 [2.6]	24	12.0 [2.6]
Role restriction	35	18.0 [5.3]	11	17.2 [5.7]	24	18.4 [5.2]	24	18.4 [3.9]
Depression	35	17.3 [3.9]	11	18 [4]	24	16.9 [3.9]	24	17.5 [5.3]
Spouse	35	16.9 [4.4]	11	18.4 [4]	24	16.2 [4.5]	24	16.9 [4.7]
Total Stress	30	210 [38.1]	10	222.2 [42.9]	20	203.9 [34.9]	24	199 [27.3]
Life Stress	35	9.4 [7.6]	11	13 [7.8]	24	7.8 [7]	24	9.46 [9.5]

One-way ANOVA with post-hoc Tukey's used for significance, * $p<0.05$ ** $p<0.01$
A= difference between SV & control
B= difference between BV & control
C= difference between SV & BV

Table 5
continued

Six Months		**All infants with CHD**		**SV**		**BV**		**Controls**
Child Domain	33	90.2 [15.6]	9	93.8 [14.4]	24	88.9 [16.1]	25	87.8 (62-112) [13.8]
Distractibility/ Hyperactivity	35	22.6 [3.9]	11	23.2 [3.7]	24	22.3 [4.1]	25	23.8 (16-31) [4.05]
Adaptability	35	22.5 [4.4]	11	22.9 [2.9]	24	22.3 [4.9]	25	22.3 [4.44]
Reinforces Parent	35	7.1 [1.5]	11	7.4 [1.9]	24	7 [1.4]	25	7.48 [1.58]
Demandingness	33	17.8 [4.6]	9	18.1 [2.9]	24	17.7 [5.1]	25	15.4 [3.22]
Mood	35	8.6 [2.2]	11	8.6 [2.1]	24	8.6 [2.4]	24	8.52 [2.76]
Acceptability	35	11.7 [3.5]	11	13.3 [3.9]*A	24	11 [3.1]	25	10.3 [2.4]
Parent Domain	35	108.5 [19.8]	11	114 [20]			24	111.3 [22]
Competence	35	23.6 [4.9]	11	25.8 [6.7]*C	24	22.6 [3.6]	25	24.4 [5.16]
Isolation	35	12.1 [4.2]	11	13.9 [4.6]	24	11.3 [3.9]	25	11.6 [3.76]
Attachment	35	10.7 [3.0]	11	11.7 [3.3]	24	10.2 [2.7]	25	11.0 [2.67]
Health	35	11.5 [2.8]	11	11.4 [2.4]	24	11.5 [3.1]	25	11.9 [2.97]
Role restriction	35	17.6 [4.2]	11	17.1 [3.2]	24	17.8 [4.7]	25	17.5 [4.4]
Depression	35	16.2 [3.9]	11	16.1 [4.2]	24	16.2 [3.8]	25	17.4 [4.55]
Spouse	35	16.9 [5.3]	11	18 [5.4]	24	16.3 [5.4]	24	17.4 [4.55]
Total Stress	33	193.3[33.3]	9	207.7 [33]	24	194.8 [33.5]	24	199 [32]
Life Stress	35	7.8 (0-23) [7.5]	11	11.8 [8.6]*A, C	24	6 [6.3]	25	5.48 [5.73]

One-way ANOVA with post-hoc Tukey's used for significance, * p<0.05 **p<0.01
A= difference between SV & control
B= difference between BV & control
C= difference between SV & BV

Table 6a. *3 Month PSI percentiles by physiology*

PSI Subscale (score range)	%ile (scores)	All infants with CHD N (%)	Infants with SV Physiology N (%)	Infants with BV Physiology N (%)	Control Infants N (%)
Distractability/ Hyperactivity	Low (9-19)	2 (6%)	--	2 (9%)	4 (16%)
	Normal (20-28)	28 (88%)	8 (80%)	20 (91%)	19 (76%)
	High (29-45)	2 (6%)	2 (20%)	--	2 (8%)
Adaptability	Low (11-19)	3 (9%)	1 (10%)	2 (9%)	2 (8%)
	Normal (20-28)	22 (67%)	6 (60%)	16 (70%)	20 (80%)
	Borderline (29)	1(3%)	--	1 (4%)	--
	High (30-55)	7 (21%)	3 (30%)	4 (17%)	3 (12%)
Reinforces Parent	Low (6)	12 (36%)	4 (40%)	8 (35%)	13 (52%)
	Normal (7-11)	18 (55%)	5 (50%)	13 (57%)	12 (48%)
	High (12-30)	3 (9%)	1 (10%)	2 (8%)	--
Demandingness*A,B	Low (9-13)	5 (16%)	1 (10%)	4 (19%)	6 (24%)
	Normal (14-21)	14 (45%)	5 (50%)	9 (43%)	18 (72%)
	High (22-45)	12 (39%)	4 (40%)	8 (38%)	1 (4%)
Mood	Low (5-6)	7 (20%)	2 (18%)	5 (21%)	5 (20%)
	Normal (7-11)	21 (60%)	7 (64%)	14 (58%)	18 (72%)
	High (12-25)	7 (20%)	2 (18%)	5 (21%)	1 (4%)
Acceptability	Low (7-8)	7 (21%)	2 (20%)	5 (22%)	8 (32%)
	Normal (9-15)	22 (67%)	6 (60%)	16 (70%)	17 (68%)
	High (16-35)	4 (12%)	2 (20%)	2 (8%)	--
Child Domain	Low (47-81)	4 (13%)	1 (10%)	3 (15%)	7 (28%)
	Normal (82-114)	22 (73%)	8 (80%)	14 (70%)	18 (72%)
	Borderline (115)	--	--	--	--
	High (116-235)	4 (13%)	1 (10%)	3 (15%)	--
Competence	Low (13-22)	18 (53%)	3 (30%)	15 (63%)	10 (40%)
	Normal (23-34)	14 (41%)	5 (50%)	9 (38%)	14 (56%)
	High (35-65)	2 (6%)	2 (20%)	--	1 (4%)
Isolation	Low (6-9)	8 (23%)	2 (18%)	6 (25%)	9 (38%)
	Normal (10-16)	24 (69%)	7 (64%)	17 (71%)	11 (46%)
	High (17-30)	3 (9%)	2 (18%)	1 (4%)	4 (17%)

Attachment *A,C	Low (7-9)	9 (25%)	2 (18%)	7 (29%)	10 (42%)
	Normal (10-15)	22 (63%)	5 (46%)	17 (71%)	13 (54%)
	High (16-35)	4 (11%)	4 (36%)	--	1 (4%)
Health	Low (5-8)	2 (6%)	1 (9%)	1 (4%)	3 (13%)
	Normal (9-15)	28 (71%)	8 (73%)	20 (83%)	20 (83%)
	High (16-25)	5 (14%)	2 (18%)	3 (13%)	1 (4%)
Role Restriction	Low (7-13)	5 (14%)	3 (27%)	2 (8%)	3 (13%)
	Normal (14-23)	25 (71%)	7 (64%)	18 (75%)	17 (71%)
	High (24-35)	5 (14%)	1 (9%)	4 (17%)	4 (17%)
Depression	Low (9-15)	10 (29%)	4 (36%)	6 (25%)	9 (38%)
	Normal (16-24)	23 (66%)	6 (54%)	17 (71%)	13 (54%)
	Borderline(25)	--	--	--	--
	High (26-45)	2 (6%)	1 (9%)	1 (4%)	2 (8%)
Spouse	Low (7-11)	3 (9%)	--	3 (13%)	4 (17%)
	Normal (12-21)	29 (83%)	10 (91%)	19 (79%)	16 (67%)
	High (22-35)	3 (9%)	1 (9%)	2 (8%)	4 (17%)
Parent Domain	Low (54-101)	11 (32%)	3 (30%)	8 (33%)	8 (33%)
	Normal (102-142)	20 (57%)	5 (50%)	15 (63%)	15 (63%)
	Borderline (143-147)	--	--	--	--
	High (148-270)	3 (9%)	2 (20%)	1 (4%)	1 (4%)
Total Stress	Low (101-187)	8 (27%)	2 (20%)	6 (30%)	8 (33%)
	Normal (188-252)	19 (63%)	6 (60%)	13 (65%)	15 (63%)
	Borderline (253-257)	1 (3%)	1 (10%)	--	--
	High (258-505)	2 (7%)	1 (10%)	1 (5%)	1 (4%)
Life Stress	Low (0-1)	6 (17%)	--	6 (25%)	3 (13%)
	Normal (2-12)	17 (49%)	5 (46%)	12 (50%)	14 (58%)
	Borderline (13)	2 (6%)	--	2 (8%)	2 (8%)
	High (14-79)	10 (29%)	6 (55%)	4 (17%)	5 (21%)

Chi-Square test of significance used, * $p<0.05$ **$p<0.01$
A= difference between SV & control
B= difference between BV & control
C= difference between SV & BV

Table 6b. *6 Month PSI percentiles by physiology*

PSI Subscale (score range)	%ile (scores)	All infants with CHD N (%)	Infants with SV Physiology N (%)	Infants with BV Physiology N (%)	Control Infants N (%)
Distractability/ Hyperactivity	Low (9-19)	7 (20%)	1 (9%)	6 (25%)	5 (20%)
	Normal (20-28)	26 (74%)	10 (91%)	16 (67%)	18 (72%)
	High (29-45)	2 (6)	--	2 (8%)	2 (8%)
Adaptability	Low (11-19)	8 (23%)	2 (18%)	6 (25%)	6 (24%)
	Normal (20-28)	26 (74%)	9 (82%)	17 (71%)	17 (68%)
	Borderline (29)	--	--	--	2 (8%)
	High (30-55)	1 (3%)	--	1 (4%)	--
Reinforces Parent	Low (6)	19 (54%)	6 (55%)	13 (54%)	10 (40%)
	Normal (7-11)	16 (46%)	5 (46%)	11 (46%)	15 (60%)
	High (12-30)	--	--	--	--
Demandingness	Low (9-13)	5 (16%)	1 (9%)	4 (17%)	7 (28%)
	Normal (14-21)	20 (63%)	7 (78%)	13 (57%)	18 (72%)
	High (22-45)	7 (22%)	1 (11%)	6 (26%)	--
Mood	Low (5-6)	6 (17%)	1 (9%)	5 (21%)	8 (32%)
	Normal (7-11)	26 (74%)	9 (82%)	17 (71%)	15 (60%)
	High (12-25)	3 (9%)	1 (9%)	2 (8%)	2 (8%)
Acceptability	Low (7-8)	8 (23%)	2 (18%)	6 (25%)	7 (28%)
	Normal (9-15)	19 (54%)	5 (46%)	14 (58%)	18 (72%)
	High (16-35)	8 (23%)	4 (36%)	4 (17%)	--
Child Domain	Low (47-81)	10 (30%)	2 (22%)	8 (33%)	7 (28%)
	Normal (82-114)	22 (67%)	7 (78%)	15 (63%)	18 (72%)
	Borderline (115)	--	--	--	--
	High (116-235)	1 (3%)	--	1 (4%)	--
Competence	Low (13-22)	14 (40%)	3 (27%)	11 (46%)	9 (36%)
	Normal (23-34)	20 (57%)	7 (64%)	13 (54%)	16(64%)
	High (35-65)	1 (3%)	1 (9%)	--	--
Isolation	Low (6-9)	9 (26%)	1 (9%)	8 (33%)	8 (32%)
	Normal (10-16)	22 (63%)	8 (73%)	14 (58%)	15 (60%)
	High (17-30)	4 (11%)	2 (18%)	2 (8%)	2 (8%)

Attachment	Low (7-9)	17 (49%)	3 (27%)	14 (58%)	8 (32%)
	Normal (10-15)	17 (49%)	7 (64%)	10 (42%)	16 (64%)
	High (16-35)	1 (3%)	1 (9%)	--	1 (4%)
Health	Low (5-8)	3 (9%)	1 (9%)	2 (8%)	2 (8%)
	Normal (9-15)	28 (80%)	10 (91%)	18 (75%)	19 (76%)
	High (16-25)	4 (11%)	--	4 (17%)	4 (16%)
Role Restriction	Low (7-13)	4 (11%)	--	4 (17%)	5 (20%)
	Normal (14-23)	29 (83%)	10 (91%)	19 (79%)	19 (76%)
	High (24-35)	2 (6%)	1 (9%)	1 (4%)	1 (4%)
Depression	Low (9-15)	15 (43%)	5 (46%)	10 (42%)14 (58%)	8 (32%)
	Normal (16-24)	20 (57%)	6 (55%)	--	16 (64%)
	Borderline(25)	--	--		--
	High (26-45)	--	--		1 (4%)
Spouse	Low (7-11)	4 (11%)	1 (9%)	3 (13%)	3 (12%)
	Normal (12-21)	25 (71%)	7 (64%)	18 (75%)	16 (64%)
	High (22-35)	6 (17%)	3 (27%)	3 (13%)	5 (20%)
Parent Domain	Low (54-101)	11 (31%)	2 (18%)	9 (38%)	7 (28%)
	Normal (102-142)	22 (63%)	8 (73%)	14 (58%)	16 (64%)
	Borderline (143-147)	1 (3%)	1 (9%)	--	--
	High (148-270)	1 (3%)	--	1 (4%)	1 (4%)
Total Stress	Low (101-187)	11 (33%)	2 (22%)	9 (38%)	10 (40%)
	Normal (188-252)	20 (61%)	7 (78%)	13 (54%)	13 (52%)
	Borderline (253-257)	1 (3%)	--	1 (4%)	--
	High (258-505)	1 (3%)	--	1 (4%)	1 (4%)
Life Stress ** A,C	Low (0-1)	12 (34%)	3 (27%)	9 (38%)	8 (32%)
	Normal (2-12)	13 (37%)	1 (9%)	12 (50%)	14 (56%)
	Borderline (13)	1 (3%)	1 (9%)	--	1 (4%)
	High (14-79)	9 (26%)	6 (55%)	3 (13%)	2 (8%)

Chi-Square test of significance used, * $p<0.05$ **$p<0.01$
A= difference between SV & control
C= difference between SV & BV

Growth. The physical growth of these infants was captured using weight, length, and head circumference. Means and Z-scores are presented below in Table 7, and significantly different z-scores between subjects and controls are noted with an asterisk. There was a significant difference between weight, length, and head circumference z-scores at three months between subjects and controls. At 3 months, infants with SV physiology weighed significantly less than control infants (p=.006). Both infants with SV and BV physiology were shorter than control infants (p=.02 and p=.046 respectively), and infants with SV physiology had head circumferences significantly smaller than control infants (p=.003). At 6 months only weight was significantly lower for infants with SV physiology compared to control infants (p=.023).

Table 7. *Descriptive statistics for Growth*

				Three months				
Growth Measure	**N**	***All infants with CHD***	**N**	***Single Ventricle***	**N**	***Bi-ventricular***	**N**	**Controls**
Weight								
Mean(kg)	35	5.52 [.92]	11	5.3 [1.1]	24	5.62 [.81]	25	6.12 [.57]
Z-score		-1.06[1.2]		-1.4 [1.4]**A		-.90 [1.1]		-.232 [.68]
Length								
Mean(cm)	35	59.3 [2.9]	11	58.7 [3.5]	24	59.6 [2.6]	25	61.6 [2.5]
Z-score		-0.83[1.2]		-1.1 [1.4]*A		-.71 [1.1]*B		.168 [1.3]
Head								
Mean(cm)	34	39.7 [1.4]	10	39.4 [1.7]	24	39.9 [1.3]	25	40.8 [1.2]
Z-score		-0.49[1.1]		-.95 [1.4]**A		-.30 [.96]		.38 [.94]
				Six months				
Growth Measure	**N**	***All infants with CHD***	**N**	***Single Ventricle***	**N**	***Bi-ventricular***	**N**	**Controls**
Weight								
Mean(kg)	35	7.26 [.98]	11	7.07 [1.3	24	7.35 [.80]	25	7.68 [.82]
Z-score		-0.76 [1.2]		-1.2 [1.5]*A		-.56 [1.0]		-.16 [.83]
Length								
Mean(cm)	35	66.3 [3.0]	11	65.8 [3.9]	24	66.5 [2.5]	24	66.6 [3.6]
Z-score		-0.58 [1.3]		-1.0 [1.7]		-.38 [1.1]		-.30 [1.4]
Head								
Mean(cm)	35	43.1 [1.3]	11	43 [1.8]	24	43.1 [1.1]	25	43.6 [1.2]
Z-score		-0.11 [1.1]		-.34 [1.3]		-.003[.97]		.437 [.96]

One-way ANOVA used for significance, * p<0.05 **p<0.01; A= difference between SV & controls; B= difference between BV & controls

Development. Table 8 presents the summary statistics for development, measured by the Bayley Scales of Infant Development-II (BSID) at six months of age. The average Psychomotor Development (PDI) score for subjects was 80.5 (50-111) and for controls was 97.3 (76-129). Infants with SV physiology scored 24 points lower than control infants (p=.000) on the PDI, and infants with BV physiology scored 14 points lower than control infants (p=.003). There was no significant difference between scores for infants with SV and BV physiology.

Table 8. *Descriptive statistics for Development (BSID-II)*

	Subjects Mean [SD]							**Controls**
	N	All infants with CHD	**N**	Single Ventricle	**N**	Bi-ventricular	**N**	
Mental Development Index	34	93.3 [8.3]	11	92.7 [6.1]	23	93.6 [9.3]	25	98.3 [7.5]
Psychomotor Development Index	35	80.5 [15.6] †A,B	11	73.7 [14.8]	24	83.6 [15.2]	24	97.3 [11.1]

One-way ANOVA with post-hoc Tukey's used for significance, † p<0.001
A= difference between SV & control
B= difference between BV & control

Specific Aim 2

Specific Aim 2 sought to examine factors of parenting stress (PS) which are associated with and predict growth (G) at 3 and 6 months of age and development (D) at 6 months of age. Birthweight was the only variable positively correlated with 3 month growth (weight- r= 0.59, p<0.001; length- r= 0.71, p<0.001; HC- r=0.54, p=0.001). Three month Role Restriction (r= -0.43, p=0.009), Spouse (r= -0.39, p=.03), and Parent Domain (r= -0.34, p=0.03) were all negatively correlated with 3 month weight z-scores. Also Hispanic infants had significantly higher weight z-scores than non-Hispanic infants (F= 1.79, p=0.032). Only 3 month Role Restriction was significantly correlated with 3 month length (r= -0.35, p=0.04). For 3 month head circumference (HC), only the 3 month distractibility mean of the EITQ was significantly correlated (r= -0.41, p=0.016). Hispanic infants had significantly larger heads than non-Hispanic infants (F= 2.24, p= 0.032).

None of the designed models predicted any of the 3 month growth outcome variables with sufficient power (≥ .80). Role restriction at 3 months and birthweight contributed to the 42% of the variance of 3 month weight z-scores (65% power). Role restriction at 3 months also contributed to 9% of the variance in 3 month length z-scores (18% power). Birthweight and length of stay contributed to 37% of the variance in 3 months HC z-scores (63% power).

For 6 month growth, the following variables were significantly correlated with 6 month weight: Birthweight (r= 0.506, p=0.002), 3 month Role Restriction (r= -0.42, p=0.01), and 6 month Isolation (r= -0.37, p=0.027); with 6 month length: Birthweight (r=0.482, p=0.003), 3 month Role Restriction (r= -0.44, p=0.008), 3 month Distractibility

mean (r= -0.365, p=0.031), and 6 month Isolation (r= -0.34, p= 0.049); and with 6 month HC: Birthweight (r= 0.426, p= 0.01). Hispanic infants had significantly larger heads than non-Hispanic infants at 6 months (F=2.44, p=0.018).

When 3 and 6 month independent variables were used in separate models to predict 6 month growth none did so with sufficient power. However, when the independent variables for the two time points were combined, there was sufficient power to predict 6 month weight and length but not head circumference. Birthweight and 3 month Role Restriction accounted for 37% of the variance in 6 month weight z-scores (84% power), and to 36% of the variance in 6 month length z-scores (86% power). Birthweight and 6 month Approach mean accounted for 24% of the variance in 6 month HC z-scores (59% power). A sample size of 35 achieved 80% power to detect an R^2 of 0.15 for weight and .14 for length attributed to Role Restriction using an F-test with a significance level of 0.05. The variables tested were adjusted for an additional one independent variable with an R^2 of 0.23 for weight and two independent variables for length with an R^2 of .30.

Predicting development was also part of Specific Aim 2. Only the Psychomotor Development Index (PDI) significantly correlated with the Mental Development Index (MDI) (r= 0.43, p= 0.01). Hispanic infants had MDI scores 10 points higher than non-Hispanic infants (F=.026, p=0.047). There were no significant predictors of the Mental Development Index scores.

The Psychomotor Development Index was correlated with 3 month Activity mean (r= 0.34, p=0.049), MDI (r=0.43, p=0.01), 3 month Role Restriction (r= -0.52, p=0.002), 3 month Spouse (r= -0.35, p=0.041), 3 month Parent Domain (r= -0.385, p=0.025), 6

month Isolation (r= -0.39, p=0.02), and 6 month Role Restriction (r= -0.56, p<0.001). Infants whose mothers graduated from college scored 14 points higher than those infants whose mothers did not have a college degree (F=.676, p=0.024). Infants who were fed by mouth (PO) at 3 months and also at 6 months scored 24 points higher than those infants who were not fed PO at 3 months or at 6 months (F=2.37, p=0.003). Using separate models for 3 and 6 month independent variables, 6 month Role Restriction and 6 month Activity mean accounted for 46% of the variance in PDI scores (91% power), and 3 month Role Restriction and 3 month Activity mean accounted for 38% of the variance (75% power). When these independent variables were combined in one predictive model, 6 month Role Restriction and 3 month Activity mean accounted for 38% of the variance (95% power). A sample size of 35 achieved 80% power to detect an R^2 of 0.18 attributed to one independent variable associated with the predictor of interest (Role Restriction) using an F-test with a significance level of 0.05. The variables tested are adjusted for an additional one independent variable with an R^2 of 0.86.

Specific aim 3

Due to the transactional nature of parent-infant relationships, Specific Aim 3 sought to understand the converse of Specific Aim 2. Its purpose was to examine factors of growth (G) and development (D) which were associated with and predicted parenting stress (PS) at 3 and 6 months.

Three month stress. The independent variables significantly correlated with 3 month Child Domain (CD) stress were: Length of hospital stay (r=0.41, p=0.025), 3 month Rhythmicity mean (r=0.37, p=0.043), 3 month Approach mean (r=0.6, p=0.001), 3 month Adaptability mean (r=0.53, p=0.002), 3 month Intensity mean (r=0.58, p=0.001), 3

month Mood mean (r=0.7, p<0.001), and 3 month Distractibility mean (r=0.63, p<0.001). Three month CD stress was not predicted by growth but by two temperament characteristics. Mood and Rhythmicity at 3 months contributed to 56% of the variance in CD stress scores at 3 months (99% power).

The variables significantly associated with 3 month Parent Domain (PD) stress were: Length of hospital stay (r=0.42, p=0.014), 3 month Rhythmicity mean (r=0.42, p=0.013), 3 month Approach mean (r=0.48, p=0.005), 3 month Adaptability mean (r=0.42, p=0.013), 3 month Intensity mean (r=0.65, p<0.001), 3 month Mood mean (r=0.51, p=0.002), 3 month Distractibility mean (r=0.43, p=.01), and 3 month weight z-score (r= -0.34, p=0.05). Parents of infants who were fed by device at 3 months had PD stress scores 36 points higher than parents of infants who were fed PO (F=3.46, p=0.002) Intensity mean temperament score and length of hospital stay accounted for 55% of the variance in 3 months PD stress scores (99% power).

The independent variables significantly associated with 3 month Total Stress (TS) scores were: Length of hospital stay (r=0.45, p=0.013), 3 month Rhythmicity mean (r=0.44, p=0.016), 3 month Approach mean (r=0.59, p=0.001), 3 month Adaptability mean (r=0.53, p=0.003), 3 month Intensity mean (r=0.68, p<0.001), 3 month Mood mean (r=0.67, p<0.001), and 3 month Distractibility mean (r=0.58, p=0.001). Parents of infants who were fed via a device at 3 months scored 51 points higher on the TS subscale of the PSI (F=0.57, p=0.023). Again, 3 month Intensity mean and length of hospital stay significantly predicted 3 month TS stress scores; they accounted for 60% of the variance with 99% power.

Six month stress. For stress at 6 months, the independent variables significantly associated with the CD were: 3 month Approach mean (r=0.47, p=0.005), 3 month Intensity mean (r=0.42, p=0.015), 3 month Mood mean (r=0.54, p=0.001), 6 month Rhythmicity mean (r=0.43, p=0.013), and 6 month Adaptability mean (r=0.48, p=0.005). When the 3 and 6 month predictive independent variables were combined in a single model, Mood and Adaptability at 3 months, Rhythmicity at 6 months and gestational age predicted 60% of the variance in 6 month CD stress (100% power). A sample size of 33 achieved 80% power to detect an R^2 of 0.08 attributed to one independent variable associated with the predictor of interest using an F-test with a significance level of 0.05. The variables tested are adjusted for an additional four independent variables with an R^2 of 0.601.

Regarding 6 month PD stress, the variables significantly correlated were: Length of hospital stay (r=0.34, p=0.044), 3 month Approach mean (r=0.51, p=0.002), 3 month Adaptability mean (r=0.45, p=0.007) 3 month Intensity mean (r=0.68, p<0.001), 3 month Mood mean (r=0.55, p=0.001), and 6 month Rhythmicity mean (r=0.39, p=0.02). When 3 and 6 month independent predictors were combined, 6 month PD was predicted by 3 month intensity and mood, maternal education, and length of stay; these variables accounted for 58% of the variance (100% power). A sample size of 30 achieved 80% power to detect an R^2 of 0.03 attributed to one independent variable associated with the predictor of interest using an F-test with a significance level of 0.05. The variables tested are adjusted for an additional four independent variables with an R^2 of 0.852.

Total Stress at 6 months was significantly correlated with: Length of hospital stay (r=0.35, p=0.047), 3 month Approach mean (r=0.54, p=0.001), 3 month Adaptability

mean (r=0.51, p=0.002) 3 month Intensity mean (r=0.61, p<0.001), 3 month Mood mean (r=0.6, p<0.001), 6 month Rhythmicity mean (r=0.44, p=0.01), and 6 month Adaptability (r=0.39, p=0.024). When 3 and 6 month predictors were combined in a single model, Intensity mean at 3 months, Adaptability mean and Rhythmicity mean at 6 months and length of hospital stay accounted for 64% of the variance in TS scores at 6 months (100% power). A sample size of 33 achieved 80% power to detect an R^2 of 0.07 attributed to one independent variable associated with the predictor of interest using an F-test with a significance level of 0.05. The variables tested are adjusted for an additional four independent variables with an R^2 of 0.658.

Specific Aim 4

Changes in stress over time. The goal of Specific Aim 4 was to identify the changes in parenting stress over the course of the first six months of life. The changes in PSI scores from 3 to 6 months were examined for all infants with CHD combined, for infants with SV and BV physiology separately, and for controls. Table 9 presents all the difference scores of subscales; the subscales with significantly different scores from 3 to 6 months are noted.

For all infants with CHD, Adaptability (2.67, p<0.001), Reinforces Parent (1.33, p=0.017), Demandingness (1.60, p=0.044), Child Domain (6.62, p=0.022), Health (.943, p=0.023), Parent Domain (3.47, p=0.042), and Total Stress (9.72, p=0.015) mean scores were significantly lower at 6 months. For infants with SV physiology, the Adaptability (3.1, p=0.025), Mood (1.55, p=0.046), Competence (2.30, p=0.034), and Depression (1.91, p=0.53) mean scores were significantly lower at 6 months. For infants with BV

physiology, two subscales were significantly lower at 6 months than at 3 months, Adaptability (2.48, p=0.007) and Reinforces Parent (1.23, p=0.038).

For controls the three subscales with significantly different scores from 3 to 6 months were Adaptability, Reinforces Parent and Life Stress. The mean difference between Adaptability scores was 1.96 (p=.005), for Reinforces Parent, -.731 (p=.054) and the Life Stress mean difference was 3.76 (p=.015). The two positive mean differences indicate Adaptability and Life Stress scores decreased from the 3 month time point to the 6 month time point, and the negative mean difference indicates the Reinforces Parent subscale scores increased from 3 to 6 months.

Table 9. *Differences in PSI subscale scores from 3 to 6 months* (3m score – 6m score)

PSI subscale	All Infants with CHD (N= 29-35)	Single Ventricle (N= 9-12)	Bi-ventricular (N= 20-24)	Controls (N=24-26)
Distractibility/ hyperactivity	.719	2.3	.000	-.346
Adaptability	2.67†	3.1*	2.48**	1.96**
Reinforces Parent	1.33*	1.6	1.23*	-.731
Demandingness	1.60*	2.89	1.05	.115
Mood	.829	1.55*	.500	-.192
Acceptability	-.758	-2.00	-.217	.000
Child Domain	6.62*	10	5.10	.846
Competence	.176	2.30*	-.708	-1.12
Isolation	.171	-.727	.583	.320
Attachment	.943	1.27	.792	-.360
Health	.943*	1.00	.917	.160
Role Restriction	.429	.091	.583	.760
Depression	1.09	1.91	.708	-.040
Spouse	.029	.364	-.125	-.333
Parent Domain	3.47*	5.20	2.75	-1.46
Total Stress	9.72*	13.9	7.85	-1.54
Life Stress	1.60	1.18	1.79	3.76*

One sample T-test used to determine significant differences *p< 0.05; ** p< 0.01, †p<0.001

Additional Analyses

Temperament as a Mediator

Infant temperament played a significant role in predicting parenting stress and growth, so further analysis was performed to determine if temperament served as a mediator in the relationship between stress and growth (Specific Aim 2) as well as the relationship between growth and stress (Specific Aim 3). There were four models which contained both temperament and the predictive independent variable. For Specific Aim 2, two models contained both a stress variable and temperament variable in the final predictive model. During the first step of testing for mediation, detecting a significant ($p<.05$) correlation between the independent and prospective mediator, there was no significant correlation detected; consequently, criteria for continuing with subsequent analysis were not met. The same was true for the two models in Specific Aim 3. It was concluded that temperament did not play a mediating role in these predictive models.

Actual Temperament vs. Perceived Temperament

The second part of the additional analyses sought to determine if the perception of infant temperament, measured by "General Impression" (GI) questions on the EITQ and ITQ was a stronger predictor than actual infant temperament, measured by the mean subscale scores. This was done by replacing the temperament means with their corresponding GI values in each of the models where temperament served as a significant predictor. In Specific Aim 2, there were four models tested, and perceived temperament did not prove to be a stronger predictor in any of the models.

In Specific Aim 3 four models were tested, and the perception of temperament was a stronger predictor than actual temperament in one. In the original model predicting

6 month PD stress, Intensity mean3 accounted for 33% of the variance in Parent Domain scores at 6 months, Maternal education for an additional 9%, Length of hospital stay for an additional 10%, and Mood mean3 accounted for an additional 6% (total variance 58%, 99.6% power). When actual mood was replaced with perceived mood, GI mood3 accounted for twice as much of the variance as actual mood (12.1& vs. 6.2%). The total variance accounted for in the model was 60% with 99.7% power. Perceived intensity and rhythmicity did not predict more than the corresponding actual temperament measures. The salient findings presented in this chapter will be discussed as well as the limitations and areas for future research in the final chapter.

Chapter 5: DISCUSSION

This dissertation sought to explore the relationships between parenting stress, growth, and development. An innovative biopsychosicial approach was used to discover previously unreported relationships. The unique knowledge this study contributes to parenting stress science is that early parenting stress does predict later growth and development. Due to the transactional nature of parent-infant interaction, temperament characteristics, namely those comprising the "difficult child constellation predict parenting stress.

This chapter will discuss in depth the findings of this novel study. The differences between subjects and controls will be discussed, as well as the differences between infants with single ventricle (SV) and biventricular (BV) physiology. The outcomes for infants with SV physiology will be highlighted due to the unbalanced share of adverse outcomes they experienced. Limitations of this study as well as theoretical and clinical implications will conclude the chapter.

Findings

Early parenting stress, specifically Role Restriction predicted later growth. The notion of psychosocial variables contributing significantly to biological outcomes has not received as much attention in the area of infant growth as other approaches, but researchers involved with biobehavioral research related to maternal and infant mental health have suggested moving away from models strictly linking growth outcomes to biological influences and moving towards models incorporating both psychological and sociological influences (Wachs, 2009). It is becoming increasingly recognized that the distinction between organic and non-organic can be artificial and some have

recommended that the evaluation of the child with failure to thrive include the concurrent assessment of the organic, the psychological, and the psychosocial (Dunne, Sneddon, Iwaniec & Stewart, 2007). Non-organic growth failure has been linked to maternal mental health variables, so perhaps the growth failure experienced by infants with CHD is not completely caused by organic issues. It might be that their growth failure is a combination of organic and inorganic factors and that is why despite surgical repair, growth failure persists for some infants. For instance, the findings of Burgess, Marshall, Rubin & Fox (2003) also support biopsychosocial models of development. They found that both early childhood temperament and parent-child relationship quality contribute to subsequent psychological/behavioral and physiological functioning. Simmons, Goldberg, Washington, Fischer-Fay, & Maclusky (1995) also found that infants with cystic fibrosis that possessed an insecure relationship with their mothers failed to improve in nutrition status and were significantly lower than healthy controls in weight for length over time. They suggest paying attention to mother-infant relationships, especially feeding interactions, as the development of more secure relationships may improve nutritional status in this population.

The finding of Role Restriction predicting later growth is original, and the mechanism through which this occurs is not fully understood. High scores on this subscale suggest that the parents experience the parental role as restricting their freedom and frustrating them in their attempts to maintain their own identity. They see themselves as being controlled and dominated by their children's demands and needs (Abidin, 1995). Role restriction has been conceptualized as one component of role strain, and functional limitations in children have been related to maternal role restriction (Silver, Bauman, &

Weiss, 1999). It is possible this role restriction interferes with a mediating process such as parent-infant attachment or maternal sensitivity and subsequently impacts infant growth. For a list of Role Restriction subscale questions see Appendix 3.

Early infant activity level and late Role Restriction predicted psychomotor development. Based on the reciprocal relationship between parents and infants, it stands to reason that more active infants would solicit more active play from their parents which in turn could increase their motor skills and abilities. After an examination of the role of parents in early motor intervention, Mahoney and Perales (2006) suggest parents are the individuals with the greatest opportunities to promote children's motor learning. Again there is a paucity of studies examining the relationship between parental stress and developmental outcomes, but Noel, Peterson and Jesso (2008) found that maternal physical stimulation was predicted by the interaction between infant temperament and mothers' reported parenting stress. Mothers of less frustrated infants provided more physical stimulation than mothers of easily frustrated infants under conditions of low-moderate stress. Mothers who reported high stress provided low levels of physical stimulation regardless of child temperament. Similarly highlighting the importance of the fit between parent and infant, Gandour (1989) explored activity level as a dimension of temperament in toddlers and found support for the hypothesis that maternal stimulation differentially influences development depending on child's activity level. This researcher suggests the importance of the match between infant activity level and stimulation level provided by the parent, and it may be that "difficult" and preterm infants have a narrower arousal range than "easy" or full term infants.

Infants whose mothers had at least a college degree had higher PDI scores. It could be that mothers with more education had more knowledge about ways to facilitate motor development or that education was a proxy for income in this sample. Also, infants who were fed by mouth had higher PDI scores than infants fed via device. Infants requiring device feedings were hospitalized significantly longer than infants fed by mouth (20 days longer). They may have consequently experienced developmental delays from the hospitalization itself or the subsequent perceived vulnerabilities an extended hospital stay fosters. When looking at the perception of vulnerability among mothers and fathers of former premature infants, Allen et al. (2004) found higher parental perception of child vulnerability (PPCV) to be correlated with lower PDI scores (but not MDI) (2004). Higher PPCV was associated with worse developmental outcomes at 1 year of age and it was predicted by maternal anxiety at discharge, which they suggest is an area amenable to intervention.

Early Parenting Stress (PS) was predicted by Mood, Rhythmicity, Intensity and length of hospital stay (LOS). In addition to these four variables, later PS was also predicted by Adaptability, gestational age, and maternal education. The "difficult child" constellation is comprised of the negative aspects of five temperament characteristics: Rhythmicity, Approachability, Adaptability, Intensity, and Mood. Four of these five characteristics predicted both early and late parenting stress. In a study of adolescent mothers of typically developing infants, Secco and Moffatt (2003) found difficult infant temperament, along with social support, were the most salient predictors of total parenting stress. Similar results were found by Saisto, Salmela-Aro, Nurmi and Halmesmaki (2008) in a longitudinal study investigating predictors of parenting stress in

mothers and fathers of toddlers. In their study, parental stress was predicted by temperament, parental characteristics such as anxiety and social support, and low self-evaluated competence. Regarding length of hospital stay, there is clear documentation supporting the stressful impact an infant hospitalization has on parents and on the newly forming parent-infant relationship (Miles & Brunssen, 2003), so the presence of this variable as a contributor to PS was expected. It is plausible that maternal education served as a proxy for income in this sample, and other studies have supported the relationship between lower SES and higher stress. Lawoko and Soares (2002) examined distress and hopelessness in parents of children with CHD, children with other diseases and other children, and found that variables such as employment status and financial situation explained more of the variance than did the disease process itself. Vilhjalmsson and Kristjansdottir found that parental role strain increases when there are two or more children in the home, parents are employed, in mothers (especially single mothers), and in lower income families (2006).

In large part the parenting stress in this study decreased over time. These findings differ from those of Uzark and Brown (2003). They found that stress increased with age. However these authors examined parents of children ages 2-12. It may be that stress during the infancy period possesses a different trajectory. They postulated that stress may increase as children age due to discipline and difficulty with limit setting. In an examination of low birthweight infants and parenting stress during early childhood, Robson (1997) found that following the infancy period the focus of parent concerns (and subsequently PS) changed to the child's developmental status and relationship patterns with the parent.

Differences

Between subjects and controls

There were interesting differences between subject and control infants regarding temperament, parenting stress and development. Subjects in the current study were less intense than control infants. This is in contrast to what has previously been described. Marino and Lipshitz (1991), in their study of infants (4-8m) and toddlers (12-36m), they found infants with CHD were more withdrawn, more intense, and had a lower threshold for stimuli than control infants. However as the infants with CHD aged, they became less active, less rhythmic, less intense, and more negative in mood than control infants.

In the current study, the temperament differences between the 3 groups that existed at 3 months were no longer present at 6 months. This is similar to what has been found in the preterm infant population. In examining the effect of intensive care exposure on temperament in a low birthweight preterm infant population, Spungen and Farran (1986) found few temperament differences between high risk preterm infants and low risk and full term infants at 6 months. They suggest that temperament may be less vulnerable to the insults of early hospitalization than physical aspects of the infants. Schraeder and Medoff-Cooper (1983) also found a similar moderating effect of temperament differences among preterm infants over time.

Regarding PS, parents of subjects experienced more stress related to their child's demandingness than parents of control infants. There were more subject mothers who reported "high" stress in this area than mothers of control infants. This is similar to what Brosig, Whitstone, Frommelt, Frisbee, & Leuthner (2007) found in that parents of infants with hypoplastic left heart syndrome (HLHS) experienced more stress on this subscale

than test norms, which made these infants more challenging to parent. High scores in the Demandingness subscale are produced when the parent experiences the child as placing many demands on him/her; these demands may come from various sources such as crying, physically hanging on the parent, frequently requesting help, or a high frequency of minor problem behaviors. In Abidin's (1995) experience, young parents tend to earn elevated scores, and parents with high scores on this subscale need guidance regarding discipline matters. In a comparison of infants with cystic fibrosis (CF), CHD, and healthy controls, Goldberg, Morris, Simmons, Fowler and Levison (1990) found though parents of infants with CHD had the highest levels of stress, parents of infants with CF rated them as more demanding.

Developmentally speaking, the subjects and controls both received scores below standardized means, but subjects scored lower than controls. Fuller et al. (2009) also found lower PDI scores in infants with CHD. Longer postoperative length of stay was predictive of lower PDI, along with suspected/confirmed genetic syndromes. These authors suggest it is infant factors such as birthweight and preoperative status rather than operative management strategies that are the most significant determinants of neurodevelopmental outcomes.

Between infants with SV physiology and the other groups

Along the continuum of outcomes, infants with SV physiology and their mothers appeared to be more adversely affected than either controls or infants with BV physiology. This is supported by what Torowicz, Irving, Hanlon, Sumpter, and Medoff-Cooper (in press) found when examining temperament and stress in a sample of infants drawn from the same parent study. They found that parenting stress was related to the

severity of the infant's CHD, in that mothers of infants with SV physiology were more stressed than mothers of infants with BV physiology. They also found that infants with SV physiology were more likely to be discharged home and maintained on multiple medications, experience multiple re-hospitalizations, demonstrate feeding difficulties, and to be at risk for profound growth failure. These sequelae coalesce to compose what the authors call the "burden of care" these families bear. Examples of the more negative outcomes for infants with SV physiology in the current study can be seen in terms of temperament, parenting stress, and growth.

Infants with SV physiology were more difficult to soothe than controls. These results are similar to those found by Torowicz et al., in which infants with SV physiology were more difficult to soothe and more negative in mood compared to infants with BV physiology and control infants (in press). Chronic and intermittent hypoxemia are associated with adverse effects on development, behavior, and academic achievement, even in children with structurally normal hearts (e.g. chronic lung disease, sleep disordered breathing, high altitude) (Wernovsky, 2006), so it may be that decreased oxygen to the brain can lead to the development of more negative behavioral style. Hughes et al. (2002) conclude temperament changes over the first year of life for preterm infants may be influenced by biological and environmental factors common to the premature birth experience.

Regarding stress, mothers of these infants experienced more competence related stress than mothers of infants with BV physiology and more than control mothers. Brosig et al. (2007) also noted parent of infants with HLHS reported higher levels of stress related to competence than parents of infants with transposition of the great arteries

(TGA). In addition, it is was found in the current study that there were more infants with SV physiology that met the "difficult child" diagnostic criteria. Others have found that infants with 'difficult' temperaments can present a greater challenge for parents and likely contribute negatively to parenting competence and stress (Secco & Moffatt, 2003).

Bithoney, Van Sciver, Foster, Corso, and Tentindo (1995) highlight the importance of the interplay between child temperament and parental sense of competence in determining growth outcomes in their study of parental stress and growth outcomes in growth deficient children. They found no differences in parenting stress between parents of children with growth deficiency and parents of controls; however, parents of children with growth deficiency (GD) perceived themselves as less competent, their children as less adaptable and reported more social isolation. A high sense of parental competence and high child adaptability were associated with improved growth outcomes. The authors felt these findings support the thesis that child adaptability and distractibility as perceived by parents may play a critical role in growth outcome and may warrant greater consideration among the range of predisposing psychosocial factors associated with GD etiology. Of special interest to them was the idea that parents with a higher sense of competence had children who grew better, suggesting parents may be able to effect positive change in their children's growth.

Mothers of infants with SV physiology experienced more early stress related to their level of attachment with their infants than either group. Others have indicated a decreased level of secure attachment experienced by mothers and their infants with CHD. Goldberg, Simmons, Newman, Campbell, and Fowler (1991) demonstrated that significantly fewer infants with CHD, in comparison with healthy peers, were considered

to have secure infant-mother relationships. Of note, the securely attached infants in this study showed greater improvements in their health than insecurely attached infants. Mäntymaa et al. (2006) reported similar findings in that poor interaction between a mother and her infant assessed as early as 2 months was associated with chronic or recurrent health problems in the child during the 2 year follow up.

High scores on the Attachment subscale suggest two possible sources of dysfunction; either the parent does not feel a sense of emotional closeness to the child, or the parent's real or perceived inability to observe and understand the child's feelings and/or needs accurately (Abidin, 1995). Either of these could be true regarding the relationship between mothers and their infants with SV physiology. Mothers of infants with the most severe CHD may feel hesitant to develop an attachment or bond with their infant due to the uncertainty of the infant's survival. The intensive care these infants often require may also limit the development of secure mother-infant relationships during hospitalization. Gardner, Freeman, Black and Angelini (1996) report difficulties in the interaction between cardiac infants and their mothers compared to non-cardiac infants. The cardiac infants had difficulty sustaining interpersonal engagement with their mothers, which caused the interaction to frequently break down. A significant number of mothers responded to the lack of engagement either by over-stimulating their infants in an attempt to regain their attention or by withdrawing and appearing distressed by their infant's behavior (which often caused the infants to withdraw further). The authors suggest difficulties in interaction stem from either the infant or the mother, and possible risk factors include: low birth weight, compromised post natal growth due to vascular disturbances such as cyanosis, and social interactions that have been hindered by

problems with breathing, eating and stamina. Though the level of engagement improved over time for the cardiac infants, it remained low when compared to non-cardiac infants. The authors suggest further investigation into ways of increasing engagement via psychological intervention.

Mothers of infants with SV physiology experienced more late stress regarding the acceptability of their infants than control parents. In exploring differences in parenting stress between parents of infants with cystic fibrosis, infants with CHD, and healthy controls, Goldberg et al. (1990) discovered parents of infants with CHD reported the highest amounts of stress, specifically related to their sense of parental competence and the acceptability of the child. Similar findings were reported by Pelchat et al. (1999), who found parents of infants with Down syndrome and infants with CHD reported significantly more stress in relation to the acceptance of their child when compared with parents of infants with cleft lip/palate and healthy controls.

High scores in the Acceptability subscale are produced when the child possesses physical, intellectual, and emotional characteristics that do not match the expectations the parents had for their child (Abidin, 1995). This was significantly higher for the parents of infants with SV physiology at 6 months, but not 3 months. As infants mature, developmental delays, especially physical ones become more evident. Perhaps the mothers of infants with SV physiology begin to realize the differences between expectations and reality as time passes because they become more pronounced or because they do not dissipate as expected. Some parents may assume the surgeries would "fix" their infants and they would be like other infants they encounter.

More infants with SV physiology were diagnosed prenatally than infants with BV physiology. Prenatal diagnosis gives parents more time to prepare for the birth of their infant with CHD, but it also gives them more time to worry and think about all the possible outcomes-good and bad. Gutteling et al. (2005) found that fear of bearing a "handicapped child" predicted the highest levels of restless/disruptive temperament and more attention regulation problems in toddlers. Brosig et al. (2007) found equivocal results when comparing parents of infants prenatally and postnatally diagnosed with CHD. Both groups of parents scored higher on the Brief Symptom Inventory than test norms at the time of diagnosis, but the scores of prenatal diagnosis group remained high 6 months after birth. In contrast, Skari et al. (2006) found that prenatal diagnosis of congenital malformations was a significant independent predictor of acute parental psychological distress after birth.

Regarding growth, as reported elsewhere (Leitch et al., 1998; Nydegger & Bines 2006), there were no significant differences in birthweight between the three groups. However, infants with SV physiology were smaller than controls at 3 months (weight, length, and HC), but had only lower weights at 6 months. Infants with CHD have been found to be prone to malnutrition and growth failure, with infants with cyanotic lesions, namely HLHS, being more affected (Varan, Tokel, & Yilmaz, 1998; Kelleher, Laussen, Tiexeira-Pinto & Duggan, 2006). Surgical correction resulted in catch-up growth in a study of 123 infants with cyanotic and congestive CHD (Schuurmans, Pulles-Heintzberger, Kester & Forget, 1998). The SV infants in the current study had only undergone two palliative surgeries at this point, so the cause of the reduction in the growth disparity is unknown. Successful cardiac surgery is usually associated with

improvements in weight within a few months, but it may take up to a year for length and HC to catch up to normal (Nydegger, & Bines, 2006).

Summary

In summary, infants with SV physiology and their parents bear the brunt of more adverse outcomes regarding infant temperament, PS and growth. Parenting stress, specifically Role Restriction, appears to contribute to infant growth and development. The mechanism is not fully understood, but perhaps it is related to the formation of secure infant-mother relationships. Also, difficult temperament characteristics, namely mood, rhythmicity, intensity, and adaptability contribute to parenting stress in this infant population.

Limitations

This study was a secondary analysis of existing data and as such had limitations. The aims of this study were not those of the parent study; consequently, this study was constrained to the questionnaires and data available. For example, the transactional nature of the phenomena studied led to the proposal of a bidirectional arrow of prediction in the theoretical model. A "chicken and egg" situation exists in that these study data did not enable to determination of which came first, the parenting stress or the growth and development challenges.

To be included in this smaller study, dyads needed complete or nearly complete data for the outcome variables; however, some incomplete data remained. It is unknown why the data were left incomplete, if the mothers did not understand the questions, if they thought the questions were not applicable or if they were accidently omitted. These would be important factors to determine if this study were replicated. Also, the PSI

contains a Marlowe-Crowne defensive responding scale that could have been used to determine if there were mothers who responded defensively and if their data should be used or interpreted with more caution. The inclusion of parents who were potentially defensive responders may have also served to alter the study findings. A defensive responding score of 24 or less indicates that the individual may be responding in a defensive manner, and caution should be exercised in interpreting that parent's scores. Extremely low total stress scores may also indicate defensive responding, but occasionally very low defensive responding scores will be found in situations where it is obvious that the parent is very competent and that the parent-child relationship exists within a supportive social situation that is economically advantaged (Abidin, 1995). To address this issue in the future, care should be exercised to evaluate potentially defensive responders on a case by case basis and make a judgment based on not just a score but on interaction with the family unit.

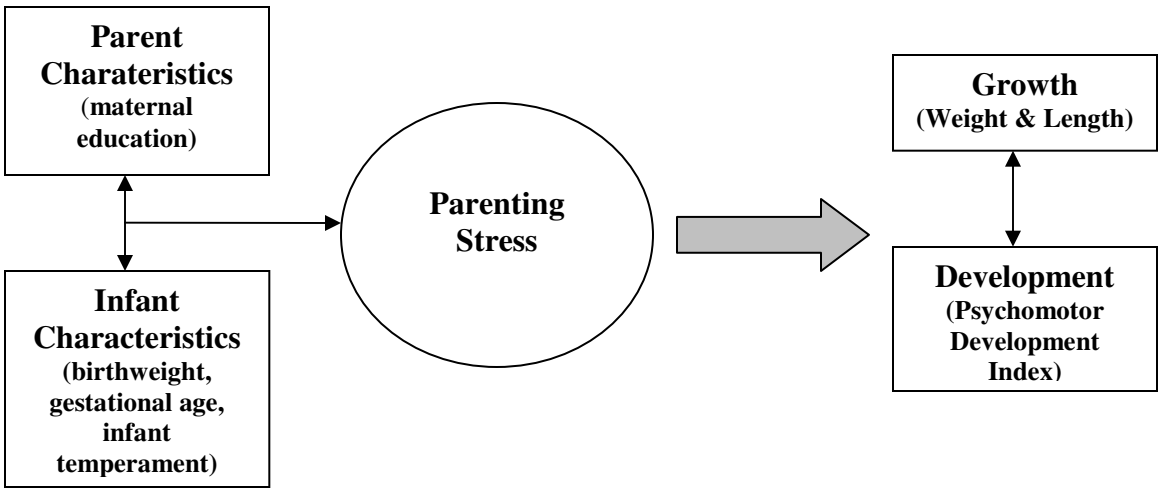
Only including infants with complete or nearly complete data decreased the sample size, which could limit the generalizability of these study findings. The fact significant results were found with sufficient power with even small sample sizes indicates the strength of the relationships tested and the importance of examining them further. The small sample did not include many infants of color; consequently, it is difficult to generalize to these populations. Though the incidence of CHD is less in African-American and Hispanic infants, their burden of illness is often times greater and these families often times experience greater stress due to socioeconomic forces, so further studies including representation from these groups would be important.

The data were collected from infants enrolled in the parent study and were part of a convenience sample from a single high volume cardiac center. This regional, national and international receiving center admits a high proportion of infants with complex congenital cardiac defects who may be at greater risk for more complicated and demanding home regimes; this may also serve to limit the generalizability of the study findings.

The mean scores on the Bayley Scales of Infant Development-II for all infants were lower than the standardized mean. This may have been due to raters underestimating the mental and motor abilities of the infants tested. The infants were tested in a new environment with a stranger. Sometimes the testing needed to be rescheduled due to an infant's sleepiness/fussiness or general lack of cooperation. Parents were often times anxious about their infant's performance, and their anxious energy may have influenced their infant's performance.

Implications

Theory/Research



Supported framework

The potential relationships between parenting stress and growth and development were presented in a proposed theoretical framework in Chapter 1. One side of the bidirectional arrow was supported while the other was not. The hypothesis that parenting stress is associated with and predicts growth and development was supported. Though maternal and infant characteristics did contribute to parenting stress, growth and development were not found to predict parenting stress. These findings support the inclusion of psychosocial variables when examining the factors that contribute to the growth and development in infants with CHD.

Parenting stress, specifically Role Restriction predicted both growth and development in infants with CHD, but the mechanism through which this occurred is not fully understood. Replication of these findings with a larger sample would help confirm the relationships found between Role Restriction and growth and development. An examination of the two remaining time points in the parent study would also yield helpful information in understanding these relationships. If Role Restriction or other subscales of the PSI remain predictive, qualitative explorations of the phenomena may further our understanding of the mechanisms behind these relationships. Other related phenomena to explore include mother-infant attachment variables and maternal sensitivity. Perhaps parenting stress alters the mother-infant relationship in some way that in turn influences infant growth and development.

Practice

The findings of this study highlight the importance of examining the "whole patient". This holistic approach to care giving is at the heart of nursing. The assessment

of parenting stress in high risk infant populations is something that is done intuitively by most nurses, but these findings illustrate the importance of continued assessment after discharge. The evaluation of stress and specifically how parents feel about their new role as full time care taker of an infant with CHD is an important dialogue to continue in the outpatient setting. Education about respite resources available and potential ways for parents to maintain their identity might prove useful in reducing the amount of role related stress these parents experience. Parents of infants with SV physiology are at particular risk and should be assessed early (even prenatally if the CHD has been diagnosed) and provided anticipatory guidance about what to expect. Education about realistic expectations and reading infant cues may also help to buffer some of the stress these families experience.

The increased stress experienced by families of infants with CHD has often been reported. This study however, provided a novel approach to examining parenting stress. The foundational knowledge gained from this study will help fuel further inquiry, and if the relationships discovered continue throughout the first year of life or can be replicated with a larger sample, a very promising area for intervention exists. Parenting stress is a mutable variable and interventions have helped reduce stress in other high risk infant populations. This nascent area of science may hold a lot of promise as researchers and clinicians try to find new solutions to old problems.

Appendix 1. Risk Adjustment in Congenital Heart Surgery-1 (RACHS-1)

Individual procedures by risk category

Risk category 1
ASD surgery (including ASD secundum, sinus venosus ASD, patent foramen ovale closure)
Aortopexy
Patent ductus arteriosus surgery >30 d of age
Coarctation repair >30 d of age
Partially anomalous pulmonary venous connection surgery

Risk category 2
Aortic valvotomy-valvuloplasty >30 d of age
Subaortic stenosis resection
Pulmonary valvotomy-valvuloplasty
Pulmonary valve replacement
Right ventricular infundibulectomy
Pulmonary outflow tract augmentation
Repair of coronary AV fistula
ASD and VSD repair
ASD primum repair
VSD repair
VSD closure and pulmonary valvotomy or infundibular resection
VSD closure and pulmonary artery band removal
Repair of unspecified septal defect
Total repair of tetralogy of Fallot
Repair of total anomalous pulmonary veins >30 d of age
Glenn shunt
Vascular ring surgery
Repair of AP window
Coarctation repair ≤ 30 d of age
Repair of pulmonary artery stenosis
Transection of pulmonary artery
Common atrium closure
Left ventricular to right atrial shunt repair

Risk category 3
Aortic valve replacement
Ross procedure
Left ventricular outflow tract patch
Ventriculomyotomy
Aortoplasty
Mitral valvotomy-valvuloplasty
Mitral valve replacement
Valvectomy of tricuspid valve

Tricuspid valvotomy-valvuloplasty
Tricuspid valve replacement
Tricuspid valve repositioning for Ebstein >30 d of age
Repair of anomalous coronary artery without intrapulmonary tunnel
Repair of anomalous coronary artery with intrapulmonary tunnel (Takeuchi)
Closure of semilunar valve, aortic or pulmonary
Right ventricular to pulmonary artery conduit
Left ventricular to pulmonary artery conduit
Repair of double-outlet right ventricle with or without repair of right ventricular obstruction
Fontan procedure
Repair of transitional or complete atrioventricular canal with or without valve replacement
Pulmonary artery band
Repair of tetralogy of Fallot with pulmonary atresia
Repair of cor triatriatum
Systemic to pulmonary artery shunt
Atrial switch operation
Arterial switch operation
Reimplantation of anomalous pulmonary artery
Annuloplasty
Repair of coarctation and VSD closure
Excision of intracardiac tumor

Risk category 4

Aortic valvotomy-valvuloplasty ≤30 d of age
Konno procedure
Repair of complex anomaly (single ventricle) by VSD enlargement
Repair of total anomalous pulmonary veins ≤30 d of age
Atrial septectomy
Repair of transposition-VSD-sub PS (Rastelli)
Atrial switch operation with VSD closure
Atrial switch operation with repair of sub PS
Arterial switch operation with pulmonary artery band removal
Arterial switch operation with VSD closure
Arterial switch operation with repair of sub PS
Repair of truncus arteriosus
Repair of hypoplastic or interrupted arch without VSD closure
Repair of hypoplastic or interrupted aortic arch with VSD closure
Transverse arch graft
Unifocalization for tetralogy of Fallot-pulmonary atresia
Double switch

Risk category 5

Tricuspid valve repositioning for neonatal Ebstein ≤30 d of age
Repair of truncus arteriosus and interrupted arch

Risk category 6

Stage 1 repair of hypoplastic left heart syndrome (Norwood operation)
Stage 1 repair of nonhypoplastic left heart syndrome conditions
Damus-Kaye-Stansel procedure

ASD, Atrial septal defect; *AV*, atrioventricular; *VSD*, ventricular septal defect; *AP*, aortopulmonary; *sub PS*, subpulmonic stenosis.

Appendix 2. *Regression models for Specific Aims 2& 3*

Full model tested	Final predictive model (R^2& adjusted R^2)			N for model	Power
SA2 Parenting Stress → Growth & Development					
(A) 3m PS → 3m G		**R^2**	**adj R^2**		
1.) 3m PS → 3m wt DV= 3m wt z-score IDV= birthweight LOS prenatal diagnosis feeding mode3 ***distractibility3 adaptability3 demandingness3 mood3*** ***role restriction3 spouse3 life stress3*** distractibility mean3	Birthweight ***Role restriction3***	.280 ***.463***	.255 ***.423***	30	.649
2.) 3m PS → 3m l DV= 3m length z-score IDV= LOS 3m feeding mode ***isolation3 role restriction3 spouse3 life stress3*** distractibility mean3	***Role restriction3***	***.120***	***.093***	35	.184
3.) 3m PS → HC DV= 3m head circumference z-score IDV= birthweight LOS prenatal diagnosis ***child domain3 competence3 attachment3 spouse3 life stress3*** persistence mean3 distractibility mean3	Birthweight Length of stay	.276 .414	.250 .369	29	.634
(B) 6m PS → 6m G					
1.) 6m PS → 6m wt DV= 6m weight z-score IDV=birthweight length of stay prenatal diagnosis RACHS1 ***acceptability6 isolation6 role restriction spouse6***	Birthweight **Isolation6**	.256 **.349**	.234 **.308**	35	.678

2.) 6m PS → 6m l DV= 3m length z-score IDV= birthweight LOS post-op physiology 6m feeding mode ***acceptability6 isolation6 attachment6 life stress6*** threshold mean6	Birthweight Threshold mean6	.233 .340	.209 .299	35	.614
3.) 6m PS → 6m HC DV= 6m head circumference z-score IDV= birthweight length of stay ***isolation6 life stress6*** approach mean6 threshold mean6	Birthweight Approach mean6	.182 .285	.157 .241	35	.591
(C) 6m PS → 6m D					
1.) 6m PS → MDI DV= Mental Development Index IDV= birthweight LOS post-op physiology gender ***competence6 isolation6 role restriction6 life stress6*** rhythmicity mean6 6m length z-score 6m HC z-score	NO SIGNFICANT PREDICTORS			--	--
2.) 6m PS → PDI DV= Psychomotor Development Index IDV= LOS feeding mode6 ***distractibility6 demandingness6 isolation6 role restriction6 spouse6*** activity mean6 6m length z-score	***Role restriction6*** Activity mean6	***.380*** .496	***.360*** .463	33	.907

(D) 3m PS → 6m G					
1.) 3m PS → 6m wt DV= 6m weight z-score IDV= birthweight LOS prenatal diagnosis RACHS1 3m feeding mode ***distractibility3 demandingness3 role restriction3 spouse3*** mood mean3 distractibility mean3	Birthweight ***Role restriction3***	.230 ***.408***	.203 ***.365***	30	.564
2.) 3m PS→ 6m l DV= 3m length z-score IDV= birthweight LOS post-op physiology 3m feeding mode ***distractibility3 isolation3 role restriction3 spouse3*** distractibility mean3	Birthweight ***Role restriction3***	.231 ***.394***	.206 ***.352***	32	.674
3.) 3m PS → 6m HC DV= 3m head circumference z-score IDV= birthweight LOS RACHS1 ***life stress3*** distractibility mean3 persistence mean3	Birthweight	.182	.157	35	.358
(E) 3m PS → 6m D					
1.) 3m PS → 6m MDI DV= 6m Mental Developmental Index IDV= LOS post-op physiology gender ***isolation3 health3 depression3*** rhythmicity mean3 3m length z-score 3m HC z-score	NO SIGNIFICANT PREDICTORS			--	--
2.) 3m PS → 6m PDI DV= 6m Psychomotor Development Index IDV= LOS ***reinforces parent3 competence3 isolation3 health3 role restriction3 spouse3*** activity mean3 rhythmicity mean3	***Role restriction3*** Activity mean3	.275 .415	.252 .376	33	.752

SA 3 Growth & Development → Parenting Stress					
(A) 3m G → 3m PS					
1.) 3m wt → 3m CD DV= 3m Child Domain IDV= LOS rhythmicity mean3 approach mean3 mood mean3 distractibility mean3 ***3m weight z-score***	Mood mean3 Rhythmicity mean3	.488 .594	.469 .564	30	.993
2.) 3m wt → 3m PD DV= 3m Parent Domain IDV= gestational age LOS post-op physiology 3m feeding mode rhythmicity mean3 intensity mean3 mood mean3 distractibility mean3 ***3m weight z-score(3m length z-score= same results)***	Intensity mean3 Length of stay	.427 .580	.409 .553	34	.992
3.) 3m wt → 3m TS DV= 3m Total Stress IDV= gestational age LOS prenatal diagnosis rhythmicity mean3 intensity mean3 mood mean3 distractibility mean3 ***3m weight z-score***	Intensity mean3 Length of stay	.456 .629	.436 .601	30	.993
4.) 3m l → 3m LS DV= 3m Life Stress IDV= maternal education ***3m length z-score (3m HC z-score= same results)***	NO SIGNIFICANT PREDICTORS			--	--

(B) 6m G → 6m PS					
1.) 6m wt → 6m CD DV= 6m Child Domain IDV= gestational age LOS prenatal diagnosis rhythmicity mean6 adaptability mean6 mood mean6 distractibility mean6 ***6m weight z-score***	Adaptability mean6 Rhythmicity mean6 Gestational age Length of stay	.228 .434 .547 .618	.203 .396 .501 .564	33	.995
2.) 6m wt → 6m PD DV= 6m Parent Domain IDV= LOS maternal education feeding mode6 adaptability mean6 approach mean6 rhythmicity mean6 mood mean6 threshold mean6 MDI PDI ***6m weight z-score***	Rhythmicity mean6 Length of stay	.156 .298	.130 .253	34	.410
3.) 6m wt → 6m TS DV= 6m Total Stress IDV= LOS gestational age rhythmicity mean6 approach mean6 adaptability mean6 mood mean6 threshold mean6 ***6m weight z-score***	Rhythmicity mean6 Adaptability mean6 ***6m weight z-score*** Gestational age Length of stay	.192 .365 ***.483*** .587 .648	.166 .323 ***.429*** .528 .583	33	.996
4.) 6m l → 6m LS DV= 6m Life Stress IDV= birthweight gestational age post-op physiology RACHS1 approach mean6 intensity mean6 mood mean6 threshold mean6 MDI ***6m length z-score***	Mood mean6 Intensity mean6 Gestational age Threshold mean6	.161 .286 .478 .565	.135 .240 .426 .505	34	.947

4a.) 6m HC → 6m LS DV= 6m Life Stress IDV= birthweight gestational age post-op physiology RACHS1 approach mean6 intensity mean6 mood mean6 threshold mean6 MDI ***6m head circumference z-score***	Mood mean6 Intensity mean6 Gestational age Threshold mean6	.161 .286 .478 .565	.135 .240 .426 .505	34	.947
(C) 3m G → 6m PS					
1.) 3m l → 6m CD DV= 6m Child Domain IDV= gestational age length of stay prenatal diagnosis approach mean3 mood mean3 persistence mean3 distractibility mean3 ***3m length z-score***	Mood mean3	.291	.268	33	.535
2.) 3m wt → 6m PD DV= 6m Parent Domain IDV= LOS maternal education feeding mode3 activity mean3 rhythmicity mean3 intensity mean3 mood mean3 distractibility mean3 ***3m weight z-score***	Intensity mean3 Maternal education Length of stay Mood mean3	.349 .456 .570 .640	.325 .416 .520 .582	30	.980
3.) 3m l → 6m TS DV= 6m Total Stress IDV= gestational age LOS activity mean3 rhythmicity mean3 intensity mean3 mood mean3 distractibility mean3 ***3m length z-score***	Intensity mean3 ***3m Length z-score***	.376 ***.486***	.356 ***.452***	33	.917

4.) 3m l → 6m LS DV= 6m Life Stress IDV= gestational age post-op physiology RACHS adaptability mean3 ***3m length z-score***	***3m Length z-score*** Post-op physiology	***.140*** .323	***.114*** .281	35	.744
4a) 3m HC → 6m LS DV= 6m Life Stress IDV= gestational age post-op physiology RACHS adaptability mean3 ***3m HC z-score***	Post-op physiology ***3m HC z-score***	.144 ***.277***	.117 ***.230***	34	.592
(D) 6m D → 6m PS					
1.) MDI → 6m CD DV= 6m Child Domain IDV= gestational age LOS rhythmicity mean6 adaptability mean6 mood mean6 distractibility mean6 ***MDI***	Rhythmicity mean6 Adaptability mean6 Gestational age Length of stay	.214 .427 .527 .600	.188 .388 .476 .540	32	.988
2.) PDI → 6m CD DV= 6m Child Domain IDV= gestational age LOS prenatal diagnosis rhythmicity mean6 adaptability mean6 mood mean6 distractibility mean6 ***PDI***	Adaptability mean6 Rhythmicity mean6 Gestational age Length of stay	.228 .434 .547 .600	.203 .396 .501 .564	33	.992

3.) MDI → 6m PD DV= 6m Parent Domain IDV= LOS maternal education 6m feeding mode rhythmicity mean6 approach mean6 adaptability mean6 mood mean6 threshold mean6 6m weight z-score ***MDI***	Adaptability mean6 Maternal education	.184 .311	.154 .258	29	.354
4.) PDI → 6m PD DV= 6m Parent Domain IDV= LOS maternal education 6m feeding mode rhythmicity mean6 approach mean6 adaptability mean6 mood mean6 threshold mean6 6m weight z-score ***PDI***	Adaptability mean6 Maternal education	.185 .312	.156 .261	30	.380
5.) MDI → 6m TS DV= 6m Total Stress IDV= gestational age LOS rhythmicity mean6 approach mean6 adaptability mean6 mood mean6 threshold mean6 6m weight z-score ***MDI***	Rhythmicity mean6 Length of stay Gestational age Adaptability mean6 6m weight z-score	.207 .366 .478 .579 .657	.181 .323 .422 .516 .591	32	.992
6.) PDI → 6m TS DV= 6m Total Stress IDV= gestational age LOS rhythmicity mean6 approach mean6 adaptability mean6 mood mean6 threshold mean6 6m weight z-score ***PDI***	Rhythmicity mean6 Adaptability mean6 6m weight z-score Gestational age Length of stay	.192 .365 .483 .587 .648	.166 .323 .429 .528 .583	33	.992

7.) MDI → 6m LS DV= 6m Life Stress IDV= birthweight gestational age post-op physiology RACHS1 approach mean6 intensity mean6 mood mean6 threshold mean6 ***MDI*** 6 m length z-score 6m HC z-score	Mood mean6 Intensity mean6 Gestational age Threshold mean6	.161 .286 .478 .565	.135 .240 .426 .505	34	.929
8.) PDI → 6m LS DV= 6m Life Stress IDV= birthweight gestational age post-op physiology RACHS1 approach mean6 intensity mean6 mood mean6 threshold mean6 ***PDI*** 6 m length z-score 6m HC z-score	Mood mean6 Intensity mean6 Gestational age Threshold mean6	.161 .286 .478 .565	.135 .240 .426 .505	34	.929

Independent predictor variables in bold italics
DV= dependent variable; IDV= independent variable

Appendix 3- Role Restriction Questions from the PSI

Answered on 5-point scale from “Strongly Agree” to “Strongly Disagree”

68. Most of my life is spent doing things for my child.
69. I find myself giving up more of my life to meet my children’s needs than I ever expected.
70. I feel trapped by my responsibilities as a parent.
71. I often feel that my child’s needs control my life.
72. Since having this child, I have been unable to do new and different things.
73. Since having this child, I feel that I am almost never able to do things that I like to do.
74. It is hard to find a place in our home where I can go to be by myself.

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