Parenting stress in parents of infants with congenital heart disease and parents of healthy infants: The first year

Introduction

Parents of infants with congenital heart disease (CHD), which is the most prevalent congenital anomaly, experience increased parenting stress, comparing to the general population [1,2]. Parenting stress, the psychological distress parents experience while trying to meet the demands of their parenting roles [3], is significant in that it focuses on the experience of parents during a potentially vulnerable time for themselves, their infants with CHD, and other family members. Infants with complex cardiac conditions (e.g. Hypoplastic Left Heart Syndrome) require early surgical interventions, followed by long-term hospitalizations in the cardiac intensive care unit (CICU). The stress their parents experience is derived from many factors, including the infant’s fragile appearance and illness complications, parenting role alteration in the CICU environment, and long separation from other family members [4,5]. Post discharge, parents face illness-related burden, increased care demands, feeding difficulties, and irritable temperaments these infants often display [1]. The increased stress has implications for family life, specifically regarding poor illness adjustment, parenting malpractices, poor quality of life, and general well-being [6,7].

The dramatic increase in complex CHD survival rates from 15% to more than 85% over the last two decades [8] means that stress during the newborn period becomes part of everyday life. Nevertheless, longitudinal assessments of parenting stress in this population have not been undertaken. The current study aimed to describe the parenting stress levels in parents of infants with complex CHD during infancy (first year of life), and compare them to the stress of healthy peers. The current study may expand our understanding regarding the stress experienced by
parents over time following the sensitive post-diagnostic/surgical period. Identification of especially stressful periods during the sensitive period of infancy may assist healthcare professionals in administering clinical protocols, educating and supporting parents, and designing stress-point interventions.

**Background**

**The Parenting Stress Model**

Parenting stress is distinct from other kinds of situational stress, and can be separately measured from other stressful circumstances [3]. The most used parenting stress model was originally developed by Abidin in 1976. Specific stress-evoking factors in the parenting role were categorized into several domains related to the parents, to the child, and to the situation [9]. Stressors in the Child Domain involve child’s temperamental and behavioral characteristics, as well as parents’ perceptions and expectations regarding their child and their parental role. These factors include the child’s adaptability (i.e. a child’s reactions to transitions), demandingness (for attention by intrusions and/or aggression), mood (as reflected by excessive crying, anxiety, or provoking anger), and distractibility/hyperactivity (which require high vigilance and active parental management). The other two factors in this domain include acceptability (how closely a child meets parental expectations), and parental reinforcement [9]. Factors constructing the Parent Domain include parents’ personality components and functionality, such as parental health, social support, attachment to the child, feelings of role restriction, and a sense of competence in the parental role. Lastly, life events occurring outside the parent–child system may moderate or exacerbate parenting stress, as they influence parental emotional resources and abilities to cope with the parenting role [9]. Child’s health was not originally included in Abidin’s model; however, the parenting stress literature provides confirmatory evidence that Abidin’s
framework can capture high parenting stress in populations coping with pediatric illness. The stress is usually captured in multiple domains, emphasizing the wide effect illness has on families.

**Parenting Stress in Populations coping with CHD**

CHD occurs in approximately one percent of live births [8]. In the United States, approximately 32,000 infants are born each year with complex, life threatening CHD conditions, requiring surgery before age one [10]. Parents of infants with complex CHD experience extremely high stress around the timing of their infant’s diagnosis, surgery, CICU stay, and home discharge [5, 11]. Families are provided with plethora of information regarding the condition, treatment, post-operative care, and symptom management, and must often make quick decisions. Depression and anxiety are common among parents, as they worry about long-term complications, and future illness implications [12]. Specifically, parenting stress is significantly higher in these parents, compared to the general population [11, 13].

The parenting stress literature reveals numerous factors that act as sources of parenting stress for parents of children with CHD. Illness severity, medical procedures, CICU hospitalizations, and increased caretaking demands are among the most common stressors in this population [1, 11, 12]. Child characteristics are also dominant stressors for parents, particularly infants’ temperamental and behavioral features such as demandingness and irritability [1, 2, 12]. Additional parental and socio-familial factors include mental and physical health, and socio economic parameters [14].

Increased parenting stress has been linked across studies to numerous adverse outcomes in both parents, and children. It has been often associated with decreased physical and psychological well-being including anxiety and depression, and poor quality of life [6,15]. Mothers are particularly vulnerable to adverse outcomes during their pregnancy, postpartum period, and
throughout the first year of infant’s life [1]. Parenting stress outcomes among children include poor social competence, and maladaptive behaviors [7, 16].

Despite the accumulating evidence of increased parenting stress in pediatric populations with CHD, this population has been scarcely investigated as compared to other pediatric populations (e.g. Autism Spectrum Disorders; Low Birth Weight infants). Additionally, studies reported on a limited ability in drawing meaningful conclusions regarding what happens to stress over the sensitive period of infancy, and have recommended on future dissemination of longitudinal research [2]. The purpose of the current study was to describe and compare parenting stress between parents of infants with complex CHD and parents of healthy infants over the critical period of infancy, during which most medical interventions occur, and during which parents adjust to the illness.

Methods

Study Design

Data for the current study was obtained from a larger prospective cohort study, conducted at a large children’s hospital in the northeast mid-Atlantic region. The study originally examined feeding aspects predicting FTT in infants with complex CHD.

Setting and Participants

A convenience sample of infants with complex CHD was recruited from the CICU. A healthy-infant sample was recruited from primary care practices. Infants included in the CHD group had complex conditions requiring corrective or palliative cardiac surgery within the first six weeks of life; were not diagnosed with other congenital anomalies or genetic syndromes (except 22q deletion and DiGeorge syndrome); were born after 35 gestational weeks, and/or
weighed >2000 grams at birth. Total enrollment for both cardiac and healthy groups was 241 infants (33% recruitment rate).

**Procedures and Data Collection**

The study was approved by the Institutional Review Board. Informed consent was signed by parents/legal guardians of all enrolled participants. Data were obtained at hospital discharge, and during four follow-up outpatient visits at three, six, nine, and twelve months of age. Infants assessments were performed by clinicians at the General Clinical Research Center, and the Nutrition and Growth Laboratory at CHOP. Parents filled out self-reporting questionnaires including demographic information, infant temperament, and parenting stress; Parents received meals and parking vouchers during visits, and gift cards upon questionnaires completion.

**Study Measurements**

**Parenting Stress Index (PSI)-Long Form.** Parenting stress was assessed at three-, six-, nine-, and twelve-month visits. The PSI is a standardized, self-reporting questionnaire designated for parents, measuring stressors in the domains identified in Abidin’s model (Child Domain, Parent Domain, and Life Stressors). The PSI-Long Form consists of 120 items, yielding scores over 17 subscales. Forty-seven items ranked on a 5-point Likert style scale (1=strongly disagree, 5= strongly agree) measure six subscales constructing the Child Domain. An example item is “My child seems to cry or fuss more often than most children”. Fifty-four, 5-point Likert style scale items, measure seven subscales constructing the Parent Domain. Item for example: “I often feel guilty about the way I feel toward my child.” Independent scores from the Parent and Child Domains are summed to an overall score, and construct the Total Stress subscale. The Life Stress Domain lists 19 stressful life events outside the parent-child system (e.g. divorce, income decrease), asking for yes/no responses. Higher PSI scores indicate higher parenting stress levels.
Interpretation of the PSI scores are mainly based on the Total Stress subscale, however, independent interpretation of the subscales’ scores reveals specific stressors in the parent-child system. The PSI was widely validated on demographically and clinically diverse samples. Alpha reliability coefficients range between .70-.90 for the different subscales [9].

Demographics and clinical parameters. Demographic and clinical parameters were obtained via parents self-reporting, and from infants’ medical records, and included parent education, race, ethnicity, infant gender, birthweight, gestational age, prenatal CHD diagnosis, illness severity, hospital length of stay (LOS), feeding mode, and infant anthropometrics.

Data Analyses

Statistical analyses were executed in STATA 13 statistical software package [17]. Descriptive statistics included measures of central tendency and variation (mean, standard deviation, median, and range) by group. Distributions of all 17 PSI subscales at each time point, for subjects and controls, were tested for normality, and for homogeneity [18]. Sub-group comparisons of parenting stress were performed (at each time point) using two-sample t-tests, to identify differences in parenting stress means at the 0.05 significance level [19]. We did not account for multiplicity, taking clinical significance based on the overall results. Power calculations were performed using PASS13 Power Calculation Software [20]. Two-sample independent t-tests were used to determine the Minimal Detectable Differences (MDD) with 80% power ($\alpha=0.05$), for all PSI subscales at each time point [21].

Findings

Table 1 displays the demographic characteristics and growth parameters of the sample. The final sample included 129 infants, mostly white ($n=102; 79\%$), non-Hispanic ($n=109; 77\%$), males ($n=84; 65\%$). The sample included 66 (51%) infants with complex CHD, of whom 34
(51%) had single-ventricle (SV) post-op physiology. The CHD infant group had a median LOS of 15 (2-159) days. Among the CHD group, 24 (37%) required device-assisted feeding (i.e. nasogastric tube, gastric tube) at discharge; and seven (10%) infants continued to require device-assisted feeding at three months of age. The CHD condition was prenatally diagnosed in 50 (75%) infants within the CHD group. Most of the mothers in the sample (62%) had college/graduate education. Groups differ on race, maternal education, and growth parameters (see Table 1).

Descriptive statistics for the PSI subscales, and PSI group comparisons are presented in Table 2. At three months of age, parents of infants with CHD had significantly higher PSI mean scores than parents of healthy controls on the Demandingness (18.61 vs. 14.95; p<0.001), Mood (9.58 vs 8.43; p= 0.024), Child Domain (95.52 vs. 88.51; p= 0.040), and Competence (24.17 vs. 22.10; p= 0.039) subscales. The Demandingness scores, experienced by parents of infants with CHD compared to parents of healthy infants, remained significantly higher throughout the entire follow-up period- at six (17.52 vs. 15.07; p= 0.006), nine (18.16 vs. 15.26; p= 0.001), and twelve (17.86 vs. 15.38; p= 0.002) months of age. Parents of infants with CHD had significantly higher mean stress scores than parents of healthy infants on the Life Stress subscale at twelve months of age (10.13 vs. 7.2; p= 0.033).

**Discussion**

The current study is the first to investigate the levels of parenting stress in population coping with CHD over the infancy period, and compare them to healthy peers. Findings demonstrate that parents of infants with CHD experience significantly higher parenting stress than parents of healthy peers on multiple subscales, and at various time points. Research over the years has reported higher parenting stress among parents of children with illnesses compared to
parents of healthy children [22, 23], and specifically in parents of children with CHD [1, 14, 24]. In some cases, the parenting stress experienced by parents of infants with CHD was even higher than those of infants with other chronic healthcare needs [13]. Our findings indicate that the CHD pediatric population may be at risk for stress-related adverse outcomes, and can benefit from further investigation of parenting stress and stress-point interventions. Early stress screening in the CICU settings may direct healthcare providers towards families at risk.

The main differences in the early stress at three months appear to be on the
Demandingness, and Mood subscales within the Child Domain, and on the Competence subscale within the Parent Domain. Per Abidin, elevated Child Domain scores usually result from child’s qualities that challenge parents fulfilling their parenting roles [9]. Specifically, elevated Demandingness scores suggest that the parents perceive the child as very dependent, or placing many demands on them. Elevated Mood scores suggest that the child displays dysfunctional affective behaviors such as excessive crying, or seems unhappy in general. Our findings resonate with other reports of increased stress on the Child Domain subscales in infants and children with CHD [2, 4]. Such studies described challenging infantile temperamental traits in infants with CHD (and in infants with other congenital defects), and particularly, irritability, intensity, difficulty to sooth, and negative moods [12, 24]. These temperamental traits were attributed to the birth and hospitalization experiences often involving hypoxemia, multiple medications, feeding difficulties, and growth failure [12, 25, 26].

The finding of higher stress on the Competence subscale in parents of infants with CHD compared to healthy controls, has been demonstrated in previous CHD studies, as well as in other pediatric populations [4, 27]. Elevated stress scores on the Competence subscale, suggest that the sources of stress may relate to parental and/or child’s characteristics (e.g. young parental
age, gaps between parental expectations and reality, child's mental/physical disability), which potentially challenge parental abilities [9]. Indeed, parents of children with CHD across studies reported difficulties in balancing parental-role functions [11, 28], and difficulties disciplining and setting limits [2]. Potentially connecting this finding to our previous finding, Secco & Moffatt suggest that the challenges presented by children’s ‘difficult’ temperaments may exacerbate feelings of parental incompetence [29]. Additionally, Author and colleagues suggested that parental perceptions regarding the child’s condition potentially affect their subjective feelings of competence as caregivers [27]. Similarly, parents of infants with CHD may feel inadequate in managing and controlling their infant’s health issues and cardiac complications. This finding is important across pediatric populations, as nurses can help caregivers reframe parental perceptions regarding the illness-related temperamental/behavioral issues, and increase parental competence with regard to illness management. Parents of infants with CHD should also be provided with adequate resources and support in the early challenging post-discharge period.

Parents of infants with CHD also experienced higher stress on the Life Stress subscale at twelve months. Elevated Life Stress scores indicate that parents must deal with stressful situational circumstances outside the parent-child system that potentially intensify the parenting stress. This stress may be due to the uncertainty of infant outcomes, or related to the ongoing parental demands such as monitoring multiple medications, ER visits, and hospitalizations during the first year. Mullen and colleagues reported close correlations between parenting stress in parents of children with CHD and their quality of life, even after controlling for disease severity [13]. Similarly, parenting stress was more closely related to family resources than to the child's health status [30]. Rimmerman and Stanger linked high parenting stress in mothers of CHD children with poor marital quality [14]. Our finding of higher Life Stress in the CHD
sample echoes other studies in the field, by demonstrating that there is a broader context of the illness implications on the family life.

**Implications**

Findings demonstrate that parents of infants with CHD generally experience higher parenting stress levels than parents of healthy infants. It might be concluded that early stress evokes around child temperamental characteristics, and around illness-related burden. Previous research highlighted the post-operational and hospital discharge periods as sensitive stressful periods for parents, as they begin to adjust and cope with the illness at home [31]. Socio-familial resources are necessary for successful parental adaptation to the CHD [10, 32], and families with fewer resources are at risk for higher distress and lower well-being [33]. The experiences, needs, and coping mechanisms of families dealing with pediatric illness are multi-faceted, and dynamic [34, 35]. (Gray, 2006; Rempel, Ravindran, Rogers, & Magill-Evans, 2013). Gray reported that early coping mechanisms in parents of children with autism include reliance on service providers and family support, whereas with time the coping strategies become more emotion-focused [34]. Similarly, Tak and McCubbin found social support to be important predictor of family coping following CHD diagnosis [36]. Early intervention should aim to promote parental adaptive coping mechanisms and productive parenting practices [33].

A recent systematic literature review indicates that the most effective interventions in reducing parenting stress were those who strived to change families’ illness perceptions and coping mechanisms [37]. Additionally, interventions that included multiple family members, and focused on specific family needs, were most efficient in promoting illness adjustment [38, 39]. Such evidence echoes stress and coping models [40]. Our findings indicate that the parenting stress emerges from multiple factors in the pediatric population with CHD, and is integrated into
various familial aspects. Therefore, stress reduction interventions should not necessarily target parenting processes exclusively [3]. Stress reduction, family-foci interventions, may have the greatest leverage for populations dealing with illness related distress. Studies suggest establishing a comprehensive and long-term system of family care, by incorporation of family-systems models into healthcare education and organizational policies [41, 42].

**Limitations and Directions for Future Research**

The sample was recruited from a single institute, and may limit the findings generalizability. The sample mostly included mothers as the primary caregivers of the recruited infants. Whereas most prior parenting stress research was conducted with samples of mothers [12, 14], studies examining both parents showed different parenting stress patterns in mothers and fathers [11]. Future research including multiple family members may contribute to the family-systems perspective [43, 44].

The PSI is a reliable general parenting stress measure validated in multiple populations, including populations with CHD. Nevertheless, recent review of the stressors across pediatric conditions demonstrated that sources of parenting stress vary per specific illness characteristics and their implications for families [45]. The authors suggested that additional illness-related factors, which are currently not included in Abidin's model, have significant impact on parenting stress [45]. More illness-specific and/or illness-related parenting stress measures exist. Utilizing such measures in clinical setting may provide information beyond those obtained from general measures [46].

Finally, the data were partially collected via self-reporting questionnaires, involving a potential risk of social desirability bias (i.e. responses are idealized to conform to socially accepted norms). However, most of the conventional research in this field utilizes self-reporting
methods, as parenting stress is mostly subjective measure of perceptions and attitudes. Lately, studies also tend to measure physiological stress as a confirmative measure to the psychological distress [47].

**Conclusions**

The current study tackled important issue in a critically ill infant population. Our findings demonstrate that parents of infants with CHD experience higher parenting stress than parents of healthy infants throughout infancy. This stress appears to result from multiple factors, such as infant’s temperamental characteristics, increased caretaking demands, parental feelings of incompetence, and general life circumstances outside the parent-child system. Family Systems interventions should be incorporated into nursing practice as high stress may put parental adjustment to the illness at risk. Early interventions following CHD diagnosis can provide mothers with immediate stress relief and time to learn and practice new skills. The intervention length and format should be carefully considered in this population, and programs should be tailored to the trajectory of parental distress and illness-adjustment.
References

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