Psychosocial functioning of parents of children with heart disease – describing the landscape

Short title: Parents of children with heart disease

What's known on this subject

- Parents of children with congenital heart disease report elevated levels of anxiety, depression and stress after cardiac surgery in infancy.
- Maternal mental health problems can have an adverse impact on the psychological adjustment of the child with congenital heart disease.

What is new

- Parents of children with milder forms of heart disease do not differ from healthy norms in the longer term and psychological outcomes are better than might be expected from early findings.
- More complex diagnoses, particularly functional single ventricle conditions and cardiomyopathy, are associated with poorer long-term psychosocial outcomes for parents.

Contributors' statements

Jo Wray conceptualized and designed the study, interpreted the findings, drafted the initial manuscript and approved the final manuscript as submitted.

Amy Cassedy carried out all statistical analyses, reviewed and revised the manuscript and approved the final manuscript as submitted.

Michelle Ernst participated in the analysis and interpretation of the findings, reviewed and revised the manuscript and approved the final manuscript as submitted.

Rodney Franklin participated in the design of the study, reviewed and revised the manuscript and approved the final manuscript as submitted.

Katherine Brown participated in the design of the study, reviewed and revised the manuscript and approved the final manuscript as submitted.

Bradley Marino conceptualized and designed the study, reviewed and revised the manuscript and approved the final manuscript as submitted.

All authors approved the final manuscript as submitted and agree to be accountable for all aspects of the work.

Abbreviations:

AHD – acquired heart disease

BV – biventricular/two ventricle

CHD – congenital heart disease

CHIP – Coping Health Inventory for Parents

FES – Family Environment Scale

HADS – Hospital Anxiety and Depression Scale

HD – heart disease

ICD – implantable cardiac defibrillator

PCQLI – Pediatric Cardiac Quality of Life Inventory

PIP – Pediatric Inventory for Parents

PTDS – Posttraumatic Diagnostic Scale

QoL – quality of life

SRRS – Social Readjustment Rating Scale

STAI – Spielberger State-Trait Anxiety Inventory

SV – functionally single ventricle

UK – United Kingdom

US – United States

Abstract

The aim was to describe the psychological functioning in parents of school-age children with heart disease (HD) in a large-scale, transnational evaluation of parent dyads across the spectrum of cardiac diagnoses and a range of psychosocial domains. Parents of children with HD attending routine out-patient cardiology follow-up visits completed questionnaires assessing their mental health, coping and family functioning. Parents (1197 mothers and 1053 fathers) of 1214 children (mean age: 12.6 years; S.D. 3.0 years; median time since last surgery: 8.9 years) with congenital or acquired HD from three centres each in the United Kingdom and the United States participated (80% response rate). Parents of children with milder HD demonstrated few differences from healthy norms and had significantly lower scores on measures of illness-related stress and post-traumatic stress than parents of children with single ventricle conditions or cardiomyopathy. Parents in these latter two diagnostic sub-groups had significantly higher levels of anxiety and depression than healthy norms but did not differ on other measures of family functioning and coping skills. There were few differences between parents from the United Kingdom and United States. Agreement between mothers and fathers within a dyad was highest for the measure of frequency of illness related stressors (ICC=0.67) and lowest for anxiety (ICC=0.12). *Conclusions*: Our results suggest two different pathways for the long-term psychological well-being of parents of children with HD: on the one hand, more complex HD is associated with poorer long-term psychosocial outcomes; in contrast there are also grounds for optimism, particularly for parents of children with less complex conditions, with better psychological outcomes noted for some groups of parents compared to previously reported early psychosocial outcomes. Future work needs to identify factors other than disease severity which might explain poorer (or better) functioning in some parents of children with more complex HD.

Introduction

Congenital heart disease (CHD) occurs in 8/1000 live births and is the most common congenital anomaly.[30] There is a wide spectrum of disease severity, ranging from minor lesions with spontaneous resolution to extremely complex, life-threatening lesions that necessitate early and multiple palliative catheter-based and surgical interventions and lifelong medical follow-up. For some children and families frequent clinic visits, hospital stays and complex medication regimens are required. The severity of the condition may necessitate family role modifications, restrict family activities and create new and considerable financial pressures, all resulting in significant parental burden. Although there have been dramatic increases in survival over recent decades, such medical and surgical advances have not been mirrored by similar increases in our understanding of the psychological outcomes of parents of children with a range of cardiac conditions.

In the general population[9], as well as in families with a child with a chronic illness[15], there is a significant relationship between parental anxiety and depression, parenting stress and family functioning and child adjustment.[2] Three recent reviews suggest that a child's diagnosis of CHD may have a significant psychological impact on their parents and that hospitalization of these children for cardiac surgery is a source of significant stress for parents[14,34,35]. However, results from these reviews were inconclusive. While elevated levels of parental stress, anxiety and depression have been found in some studies, particularly those of parents of infants who had recently undergone surgery,[1,10,29,18] parents in other studies had levels of psychological functioning which were comparable to parents of healthy children or published norms.[32,31,25] Study variability in domains evaluated and measures used, the time at which parents were assessed, sample sizes and the inclusion of fathers as well as mothers is likely to explain some of the disparate findings. There is evidence that maternal mental health problems can have a negative impact on the child with CHD,[25,31,22] reinforcing the importance of assessing parental psychosocial functioning in the provision of holistic care to the child and

family. A number of medical and psychological factors have been identified as correlates of parental adjustment–for example, time since cardiac surgery and the number of cardiac operations, family cohesion and family conflict[4,27,6,32]–but, as has been found in other illness conditions, the relationship between parenting distress, depression and anxiety, and disease severity is less clear.

While there is a growing body of evidence about the psychological impact of CHD on parents, particularly during infancy, far less is known about the impact on parents when children have an acquired or inherited heart condition (e.g. cardiomyopathy or electrophysiological conditions). Compared with CHD there is greater variability and less stability over time in the incidence and presentation of different acquired or inherited conditions. In some cases children who are seemingly completely healthy can deteriorate quickly, with all of the attendant stress and anxiety for parents, and their condition may become life-threatening, necessitating invasive treatments such as the implantation of a pacemaker or cardiac defibrillator (ICD) or heart transplantation. Limited data suggest parents of children with or at risk of long QT syndrome have elevated levels of fear and anxiety[7,11] and quality of life (QoL) of children and adolescents with an ICD[5] was correlated more strongly with family functioning than with the severity of their heart condition, indicating the importance of assessing factors other than just clinical severity. There are few published data about parental and family functioning in these rarer diagnostic groups, thus the need for an overview of the 'landscape' of psychological functioning of parents of school-age children with heart disease (HD) – i.e. a large scale, international evaluation of parental (parent-parent) dyads across the spectrum of cardiac diagnoses and a range of psychosocial domains. This study addresses this gap in the existing literature. Specifically, the purpose of this study was to:

1. Determine whether a large population of parents of children/adolescents with HD differ significantly by disease complexity and from published normative values.

2. Examine whether there are differences in parental responses between parents in the United Kingdom (UK) and parents in the United States (US).
3. Determine the level of agreement between parental dyads, testing whether mothers significantly differed from fathers.

Methods

Study Design

The Pediatric Cardiac Quality of Life Inventory (PCQLI) Testing Study was a multi-center, cross-sectional study which involved the testing of a disease-specific QoL measure for pediatric patients with HD at ten tertiary hospitals in the UK and US.[36,20] For this corollary study, a subset of parents at six of the sites (three each in the UK and US) also completed measures of their own psychological functioning.

Study Population and Data Collection

Parents of children aged 8-18 years with a known heart condition (congenital or acquired) were included in the PCQLI Testing study (consent rate: 85%). Children with a genetic syndrome or learning disability (and their parents) were excluded if the children themselves were unable to complete questionnaires. Children due to attend a routine outpatient cardiology appointment were identified from clinic lists and families were contacted before their appointment to inform them about the study. Families who agreed to participate were mailed questionnaires (for both parents if appropriate) prior to their clinic appointment and asked to complete and bring them when they came to clinic. On the day of the clinic parents were asked to complete some additional, paper questionnaires prior to seeing the physician. If two parents attended clinic they were both asked to complete clinic questionnaires although frequently only one parent attended.

To detect parental differences that may be due to severity of HD, pediatric patients were classified based upon their heart condition as follows: 1) two-ventricular unrepaired CHD; 2) two-ventricle lesions with previous biventricular (BV) heart surgery and/or catheter-based intervention; 3) functionally single-ventricle (SV) with previous surgery and/or catheter-based intervention; 4) cardiomyopathy; 5) pacemaker or ICD and 6) acquired heart disease (AHD). These categories were mutually exclusive but while BV participants with a pacemaker were classified as pacemaker patients, SV participants with a pacemaker were classified as SV. We believe that SV patients with and without pacemakers are affected by their heart complexity despite the presence or absence of a device. Clinical information collected from the medical notes included previous hospitalizations, number of surgeries and interventional/diagnostic catheters and medications. Parents provided demographic information about their race, employment and education.

Measures

A range of psychosocial factors common to many models of psychosocial adaptation to chronic illness were assessed[33,28] – specifically anxiety (Spielberger State-Trait Anxiety Inventory[24] and Hospital Anxiety and Depression scale),[37] coping (Coping Health Inventory for Parents),[21] stress (Post-traumatic Diagnostic Scale,[8] Social Readjustment Ratings Scale[12] and Pediatric Inventory for Parents[26]) and family functioning (Family Environment Scale).[23] Selection of measures took into account their use in previous research, participant burden and time available for completion (particularly in clinic). Details of questionnaires are provided in Table 1.

Statistical Analysis

Summary statistics for continuous variables were expressed as means or medians and standard deviations (confidence intervals, where appropriate) or ranges. Categorical variables were expressed as percentages. Socio-demographic differences were tested using χ^2 or t-tests. To

determine between-diagnostic group significance, logistic regression (nominal data) and analysis of covariance (continuous data) was used, controlling for study site. A Bonferroni correction, for multiple group comparisons, was used for this series of analyses with the corrected probability reported. T-tests and χ^2 were calculated to compare published normative data, where available, and diagnostic groups. Parental gender and country comparisons were tested using either logistic regression or analysis of covariance, controlling for study site and diagnostic group. Levels of agreement between parents within a dyad were assessed using a weighted Kappa (nominal data) and Interclass Correlation Coefficients (ICC) (continuous data). Models included study site and diagnostic group. An alpha level of 0.05 was considered statistically significant and data was analyzed using SAS 9.2©.

The Ethics committees and Institutional Review Boards of all participating institutions approved the study. Written (UK) or verbal (US) consent was obtained from all participating parents attending clinic. For parents who completed questionnaires at home only, completion was presumed to imply consent.

Results

Parents of 1214 children (composed of 1036 dyads and 178 single parents) participated in this project, representing 80% of those approached from the PCQLI Testing study. There were no differences in demographic variables between those parents who consented to participate and those who did not consent. Table 2 shows the demographic information for the 1197 mothers and 1053 fathers completing questionnaires. Eighty-five percent (n=1027) of primary caregivers were mothers and 15% (n=187) were fathers. The US sample had significantly more White/Caucasian parents compared to the UK sample ($p<.001$). Finally, consistent with between-country educational differences[13], significantly more US parents from the US graduated from College/University ($p<.001$).

Medical and demographic data for the children are shown in Table 3. More US patients had not had any hospital stays (due to the higher proportion of children with mild CHD or acquired HD from the US sites) and more UK patients had had 11 or more cardiac-related hospitalizations, due to the higher proportion of SV patients from the UK sites. There were also some significant country differences with respect to patients in educational programs and medical care utilization.

Comparisons by disease complexity/diagnostic group

Supplemental Table 1 provides results for mothers and fathers in each diagnostic group. Overall there was a trend for parents of children with SV conditions or cardiomyopathy to have poorer psychological functioning than parents in the other groups, particularly parents of children with mild CHD or BV conditions. Parents of children with pacemakers also had scores indicative of poorer functioning on several measures when compared with parents of children with mild CHD. Significant differences between the groups for both mothers and fathers were primarily on the measures of post-traumatic stress and stress related to coping with the child's condition. Mothers and fathers of children with mild CHD also used the coping strategy of 'understanding the medical situation through communication with other parents and medical staff' less than parents of children in the other groups.

Comparisons with normative values

Comparisons with normative values indicated relatively few differences (Supplemental Table 1). Parents of children with SV conditions, cardiomyopathy, or pacemakers, had higher scores on the HADS anxiety than the normative population. There were also some differences on the Family Environment Scale, with HD groups having higher levels of cohesion, expressiveness, organization and control, and lower conflict compared with normative values.

Gender-by-country comparisons

Gender-by-country comparisons on the individual measures revealed few differences between the UK and US. Mothers in the US [n=78 (14.3%)] were significantly more likely to report having a 'Moderate' to 'Major' Social Readjustment compared to their UK counterparts [n=34 (5.8%)], ($\chi 2$=25.12; p<0.001), while UK mothers [n=205 (40.6%)] were more likely to report higher levels of anxiety on the STAI compared to US mothers [n=156 (32.4%)], ($\chi 2$=4.75; p=0.029). US mothers [Mean = 18.5 (CI: 17.9-19.1) reported higher levels on role frequency on the PIP compared to UK mothers [Mean = 17.6 (CI: 16.8-18.3)] (f=4.61, p=0.032). US fathers reported higher levels of organization [Mean = 51.5 (CI: 50.1-52.9)] compared to UK fathers [Mean = 49.3 (47.7-50.90], (f = 5.26, p = 0.022). Both mothers and fathers in the US reported higher use of all three coping strategies on the CHIP compared with parents in the UK (Table 4). A similar pattern was seen on the Family Environment Scale Control subscale.

Parental agreement within a dyad

Dyads had the highest level of agreement on the Pediatric Inventory for Parents, Total Frequency score (ICC=0.67) and the lowest level of agreement on the Spielberger State-Trait Anxiety Inventory (STAI) (ICC=0.12). Comparison of mothers' and fathers' scores indicated mothers had higher levels of anxiety (HADS-Anxiety) and illness-related stress (STAI), but were more likely to report higher levels of family expressiveness (FES) and organization (FES) than fathers (Table 5).

Discussion

This is the first large-scale study of which we are aware to describe psychological functioning in parents of school-aged children with congenital or acquired HD. More than 2200 parents were included and, in contrast to previous studies of parental adjustment, we also recruited a large sample (n=1053) of fathers. This sample size gave us the ability to test for differences

between HD groups as well as examine between-country differences for parents. In addition we were able to examine the level of agreement within parental dyads.

Our study results provide a positive message about the mental health and adjustment of parents of school-aged children with milder heart conditions in particular, with few reported differences compared to normative populations. Parents of children with single ventricle and cardiomyopathy had significantly higher levels of anxiety and depression than healthy norms but did not differ on other measures of family functioning and coping skills. There were relatively few differences between parents in the UK and US, despite differences in health service delivery and welfare state provision, supporting the potential generalizability of our findings to other Western countries. However, these differences in health service and welfare state support may explain the main differences that did exist between parents in the two countries, which were the greater use of adaptive coping strategies and higher levels of control by parents in the US. The free-at-the-point-of-delivery health care and easier access to state-provided support in the UK may result in UK families relying to a greater extent on external sources of support rather than developing and using resources within themselves and their family. While the level of agreement between mothers and fathers within a dyad was significant for the total sample on all measures of psychological functioning, mothers were more anxious and reported more illness-related stress than fathers.

In contrast to much of the existing research which focused on infancy and the early post-operative period and described elevated levels of mental health problems and psychological distress in parents of children with CHD, our data describe functioning of parents of older children and adolescents, some of whom were many years out from cardiac surgery or had diagnoses such as cardiomyopathy which have been less frequently studied. Importantly, the longer-term psychological outcomes are better for some groups of parents of children with HD than might be expected from reported early results. Our findings support those of a smaller

study of parents of school-aged children who had undergone repair of less complex lesions at least 7 years previously, reporting lower levels of distress and similar coping styles compared with controls.[25]

In agreement with some previous findings,[35,14] parents of children with more complex HD had poorer psychological adjustment than healthy norms or parents of children with less severe HD. Children with SV conditions in particular are likely to have undergone multiple surgeries, frequent hospitalizations, and require more frequent medical follow-up and daily medications than their counterparts with less severe HD. Such requirements often place significant caregiving burdens on parents, with a resulting impact on their own psychological adjustment. Furthermore, the uncertainties associated with long-term outcomes for children with SV conditions are likely to be an additional source of stress and anxiety to parents, coupled with managing their child's own reactions to their heart condition and requirement for monitoring and medical interventions. Interestingly, parents of children with cardiomyopathy were more similar to parents of children with SV conditions than to other cardiac groups. There is little published about the coping and adjustment of parents of children with cardiomyopathy but the severity, impact and uncertainty associated with cardiomyopathy – and in particular the potential for rapid, life-threatening deterioration and requirement for invasive, urgent treatments – are likely to contribute to levels of stress and anxiety.

Family levels of cohesion were higher and levels of conflict lower than published norms, suggesting a pattern of resilience and adaptation. Levels of organization and control were also higher than published norms, suggesting that planning and rules are used to organize family life. In a recent systematic review[19] significant positive correlations were identified between family cohesion, organization and lack of conflict and children's psychological health, social competence and behavior across a range of chronic illnesses, including CHD.[16] Our findings are therefore encouraging for the psychological wellbeing of children with HD.[3]

Mothers generally had higher levels of anxiety and symptoms of post-traumatic stress and illness-related stress than fathers, supporting previous findings.[17,25] Mothers are more likely than fathers to assume the primary care-taking role, including responsibilities related to their child's heart condition. Mothers are also more likely to experience social isolation as a result of their child's heart condition and may have had to forego their own careers, particularly if their child had more complex HD, which may have resulted in less social support and a greater focus on the requirements of managing their child's heart condition. However, within parental dyads there was agreement on all measures of psychosocial functioning, suggesting that parents may intentionally or unintentionally coordinate their response to their child's heart condition.

Findings from this study have implications for the delivery of psychosocial interventions. Firstly, it is evident that parents of children with HD require routine screening to enable appropriate referral to psychosocial services to address their needs and enhance their ability to cope with the myriad of stressors they encounter. Secondly, building on identification of similarities and differences in different domains of psychosocial functioning between different groups of parents will enable a more nuanced understanding and characterization of factors which enhance adaptation, which in turn can inform service provision. Thirdly, evaluation of the impact of psychological, mental health and social work services will further facilitate effective and efficient targeting of services – for example, the positive outcomes for some groups in our study may be due to earlier psychosocial intervention but without a systematic approach to evaluation we cannot say what works for whom and why.

There are some limitations to the current study, primarily related to missing data and the fact that some parents completed home questionnaires but not clinic questionnaires whereas other

parents only completed clinic questionnaires. Furthermore, often only one parent came to clinic. Although the overall study sample was large, some sample sizes and number of parental dyads in specific diagnostic groups were less than 100, but this still compared favorably with the published literature. Additionally, the choice of measures meant normative data did not exist for all measures. The proportion of academically gifted children in the US sample was high and the proportion of children with a significant mental health problem was low which may limit the generalizability of our findings to other groups of children with HD, although it is important to note that these data were provided by parents and not independently verified. Finally, the US and UK populations did differ in terms of HD diagnosis and resultant medical history, with a higher proportion of SV patients and a lower proportion with mild CHD and acquired HD from the UK sites, potentially reflecting some differences in the heart programmes at the individual sites.

Conclusion

Our results suggest that there are two stories about the long-term psychological wellbeing of parents of children with HD. On the one hand there are grounds for optimism for parents of those with less complex conditions, with potentially positive consequences for the children themselves. However, it is also evident that some parents – particularly those whose children have more complex CHD or cardiomyopathy - fare worse than others and we now need to understand what factors may be linked to poorer (or better) psychosocial outcomes. Although within the group of parents of children with more complex CHD or cardiomyopathy a proportion of parents do well it is not clear what protective factors might explain this. In the context of findings for infants and younger children, our results emphasize the importance of longitudinal psychosocial screening for all parents of children with HD and further research is now needed to identify factors, other than disease severity, which may increase the risk for poorer psychological outcome. Understanding how disease severity and other factors impact psychosocial outcomes, and whether any parental factors moderate or mediate the relationship

between clinical variables and psychological outcomes and quality of life of children with HD, is the next step to inform the implementation of targeted interventions to improve psychosocial outcomes.

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Compliance with ethical standards

Potential Conflicts of Interest

Bradley S. Marino, MD, MPP, MSCE is the creator of the Pediatric Cardiac Quality of Life Inventory. The authors do not have any other conflicts of interest relevant to this article to disclose

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Ethical approval

Appropriate ethical and institutional review board approval was received at all sites.

Informed consent

Parents provided written (UK) or verbal (US) consent for their participation.

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Table 1: Questionnaires completed by parents

Questionnaire	Domains measured	Description	Time to complete (minutes)	Place of completion
Spielberger State-Trait anxiety inventory (STAI)	Anxiety	20 items; questions rated on 4 point scale; higher scores indicate greater anxiety; scores range from 20-80	10	Clinic
Hospital Anxiety and Depression (HADS) scale	Anxiety and Depression	14 items; 2 subscales measuring anxiety and depression, 7 items each; total scores range from 0-21 on each subscale; scores of 0-7 are considered normal; scores of 8-10 indicate mild, 11-14 moderate and >15 severe levels of anxiety or depression;	5	Home*
Coping Health Inventory for Parents (CHIP)	Coping	45 items; assesses parents' perceptions of the helpfulness of behaviors they utilize to manage family life when they have a seriously or chronically ill child; three coping strategy subscales: maintaining family integration, cooperation and an optimistic definition of the situation; maintaining social support, self-esteem and psychological stability; and understanding the medical situation through communication with other parents and consultation with the medical staff; higher scores indicate that a behavior is more helpful	10	Home
Post-traumatic Diagnostic Scale (PTDS)	Post-traumatic stress	Scale modified for this study to relate specifically to the diagnosis of child's heart condition (rather than to any traumatic event); parents asked whether they felt their child's life was in danger (yes/no) and whether they felt intense fear, horror or helplessness (yes/no) when their child's heart condition was diagnosed; given a list of 17 items corresponding to DSM-IV PTSD symptoms of re-experiencing (5 items), avoidance (7 items) and arousal (5	10	Clinic

		items) and asked to rate frequency of each symptom in the past month on a 4 point scale (0=not at all or only one time to 3=five or more times a week/almost always); indicated whether or not any of the 17 symptoms had impacted on their daily life in 9 domains (e.g. work, relationships with family, fun and activities) during the previous month; total symptom severity score could range from 0-51.		
Family Environment Scale (FES)	Family functioning	2 scales of FES used: Family Relationship and System Maintenance; 45 questions with 5 subscales measuring family cohesion, expressiveness, conflict, organization and control; scores on each subscale range from 0-9; higher scores indicate greater endorsement of the dimension	15	Home
The Social Readjustment Rating Scale (SRRS)	Life stress	43 items considered to be life stressors, each with a pre-assigned value of 'life changing units' reflecting relative amount of stress caused by the event; asked to indicate which, if any, of the events have happened in previous 12 months; total score calculated taking account of weighting for each item; higher scores denote greater life stress; scores of 0-149 correspond to little life stress, 150-199 mild, 200-249 moderate, 250-299 serious and >300 serious life stress.	10	Home
Pediatric Inventory for Parents (PIP)	Illness related stress	42-items; for each item parents report frequency and difficulty with which they experience stress related to that element of caring for their child as a result of their illness; four domains: communication, emotional functioning, medical care and role functioning; two total scores reflect *frequency* of stressful events and degree of *difficulty* experienced by parents in coping with these events, with higher scores indicating greater frequency and difficulty and thus higher levels of parenting stress.	15	Home

*The HADS was introduced part-way through the study and was therefore not available to all parents to complete

Table 2: Parent Characteristics

Table 2. Parent Characteristics								
	Mothers				*Fathers*			
Variable	Total	UK	US	p Value	Total	UK	US	p Value
	N = 1197	624 (52.1)	573 (47.9)		N = 1053	545 (52.0)	506 (48.0)	
Caregiver Status, n (%)				0.987				0.383
Primary Caregiver – single parent	161 (13.5)	83 (13.3)	78 (13.6)		17 (1.6)	6 (1.1)	11 (2.2)	
Primary Caregiver in a Dyad	866 (72.4)	452 (72.4)	414 (72.3)		170 (16.1)	89 (16.3)	81 (16.0)	
Secondary in a Dyad	170 (14.2)	89 (14.3)	81 (14.1)		866 (82.2)	452 (82.6	414 (81.8)	
Age, mean (SD)	43.1 (6.4)	42.9 (5.9)	43.3 (6.8)	0.212	45.5 (6.7)	45.8 (6.5)	45.3 (6.8)	0.2785
Race, n (%)				< 0.001				< 0.001
Asian	74 (6.3)	62 (10.0)	12 (2.2)		63 (6.2)	55 (10.3)	8 (1.6)	
American Indian/Pacific Islander	3 (0.2)	0 (0.0)	3 (0.5)		3 (0.3)	0 (0.0)	3 (0.6)	
Black or African-American	74 (6.3)	28 (4.5)	46 (8.2)		47 (4.6)	16 (3.0)	31 (6.4)	
White	1018 (86.3)	521 (84.0)	497 (88.9)		902 (88.2)	458 (85.6)	444 (91.0)	
Mixed	10 (0.8)	9 (1.5)	1 (0.2)		8 (0.8)	6 (1.1)	2 (0.4)	
Hispanic, n (%)	17 (1.4)	4 (0.6)	13 (2.3)	0.017	20 (1.9)	2 (0.3)	18 (3.5)	<0.001

Highest Educational Attainment Level, n (%)				< 0.001				< 0.001
Less than High School (or Equivalent)	29 (2.4)	7 (1.1)	22 (3.8)		28 (2.7)	3 (0.6)	25 (5.0))	
High School Graduate (or Equivalent)	440 (37.0)	316 (51.3)	124 (28.2)		382 (37.1)	265 (50.5)	117 (23.2)	
Some College (or Equivalent)	288 (24.2)	141 (22.9)	147 (25.7)		219 (21.3)	113 (21.5)	106 (21.0)	
College (University) Graduate	308 (25.9)	108 (17.5)	200 (35.0)		259 (25.2)	108 (20.6)	151 (29.9)	
Post Graduate Degree	123 (10.3)	44 (7.1)	79 (13.8)		142 (13.8)	36 (8.9)	106 (21.0)	
NOTE: Column percentages are reported								

Abbreviations: US – United States; UK – United Kingdom

Table 3: Child/Adolescent Characteristics

	Total	UK	US	p value
	N = 1214	630 (51.9)	584 (48.1)	
Demographic				
Male, n (%)	663 (54.6)	339 (53.8)	324 (55.5)	0.5593
White, n (%)	1018 (85.2)	519 (82.9)	499 (87.5)	0.1471
Hispanic, n (%)	23 (1.9)	1 (0.2)	22 (3.8)	< 0.0001
Age (years), mean (SD)	12.6 (3.0)	12.6 (2.8)	12.8 (3.2)	0.2414
Live with both parents, n (%)	897 (73.9)	457 (72.5)	440 (75.3)	0.2666
Number of children < 18 (including patient) live in the household, median (range)	2 (1 - 8)	2 (1 - 8)	2 (1 - 8)	0.5171
Medical History				
Child was premature, n (%)	174 (14.4)	86 (13.7)	88 (15.1)	0.48
Age (years) child was diagnosed with heart disease, n (%)				< 0.0001

Prenatal	125 (10.3)	75 (11.9)	50 (8.6)	
Pre-discharge from the newborn nursery	379 (31.2)	200 (31.7)	179 (30.7)	
Post-discharge to 29 days of life	121 (10.0)	84 (13.3)	37 (6.3)	
30 days of life to 1 year	221 (18.2)	139 (22.0)	82 (14.0)	
> 1 year of life	366 (30.1)	131 (20.8)	235 (40.2)	
Heart Complexity (mutually exclusive categories), n (%)				< 0.0001
Mild CHD (two-ventricle unrepaired CHD)	210 (17.3)	68 (10.8)	142 (24.4)	
Two-ventricle with S/p heart surgery (not transplant) and/or catheter based intervention	544 (45.0)	332 (52.9)	212 (36.4)	
Single-ventricle S/p heart surgery (not transplant) and/or catheter based intervention	130 (10.7)	95 (15.1)	35 (6.0)	
Cardiomyopathy	69 (5.7)	43 (6.9)	26 (4.5)	
Pacemaker or ICD	101 (8.3)	46 (7.3)	55 (9.5)	
Acquired heart disease	156 (12.9)	44 (7.0)	112 (19.2)	
Child was diagnosed with a stroke, n (%)	22 (1.8)	15 (2.4)	7 (1.2)	0.1364
Child was diagnosed with a seizure disorder, n (%)	22 (1.8)	8 (1.3)	14 (2.4)	0.1956

Child was diagnosed with a learning disability, n (%)[a]	216 (17.8)	113 (17.9)	103 (17.6)	0.8916
Child was diagnosed with a significant mental health problem, n (%)[a]	21 (1.7)	6 (1.0)	15 (2.6)	0.0451
Medical Care Utilization				
Hospitalizations				
Cardiac-related hospitalizations, n (%)				< 0.0001
0 times	315 (26.0)	76 (12.1)	239 (40.9)	
1 time	314 (25.9)	174 (27.6)	140 (24.0)	
2 - 5 times	382 (31.4)	227 (36.0)	155 (26.5)	
6 - 10 times	118 (9.7)	90 (14.3)	28 (4.8)	
11 - 20 times	46 (3.8)	36 (5.7)	10 (1.7)	
> 20 times	38 (3.1)	27 (4.3)	11 (1.9)	
Time (years) since last hospitalization, median (range)	5.4 (0 - 18)	5.2 (0 - 17.8)	5.8 (0 - 18)	0.4008
Number of cardiac surgeries, median (range)	1 (0 - 7)	1 (0 - 7)	0 (0 - 6)	< 0.0001

Time (years) since last surgery, median (range)	8.9 (0.1 - 18)	9 (0.1 - 18)	8.9 (0.1 - 18)	0.6554
Number of cardiac catheterizations, median (range)	1 (0 - 20)	1 (0 -10)	0 (0 - 20)	< 0.0001
Time (years) since last catheterization, median (range)	5.2 (0 - 17.9)	5.5 (0.1 - 17.9)	4.8 (0 - 17.3)	0.0281
Number of doctor's visits in past 12 months, median (range)	3 (0 - 40)	2 (0 - 25)	3 (0 - 40)	< 0.0001
Number of medications, median (range)	0 (0 - 19)	0 (0 - 10)	1 (0 - 19)	0.1242
Education[a]				
Educational Programs (check all that apply), n (%)				
Gifted program in school	147 (13.4)	59 (9.4)	88 (18.8)	< 0.0001
Self-contained special education classroom (full-time)	19 (1.8)	12 (1.9)	7 (1.5)	0.8155
Some special education classes (not full-time) and some mainstream classes	53 (4.8)	20 (3.2)	33 (7.1)	0.004

Some special education classes (not full-time) without mainstream classes	10 (0.9)	3 (0.5)	7 (1.5)	0.1053
Learning supports (tutor or learning disability services)	128 (11.7)	82 (13.0)	46 (9.9)	0.1063
Child has an individualized educational plan	138 (12.6)	62 (9.8)	76 (16.4)	0.0016
Number of missed school days in the past year, median (range)	3 (0 - 97)	3 (0 - 97)	3 (0 - 49)	0.054

[a] Information collected directly from parents

Table 4: Parental scores on instruments, agreement between parents, and tests for differences

Mothers		**UK**	**US**	**F Value**	**Probability Value**
Coping Health Inventory for Parents (CHIP), Least Square Mean (CI)#	**Coping I: Family integration, cooperation and optimistic definition of situation (n = 954)**	36.4 (35.0-37.9)	41.3 (40.0-42.5)	30.81	<0.001
	Coping II: Social support, self-esteem, and psychological stability (n = 948)	25.4 (24.0-26.8)	28.5 (27.2-29.7)	13.03	<0.001
	Coping III: understanding medical situation thru communication with other parents and medical staff (n = 567)	13.6 (12.9-14.3)	14.8 (14.2-15.4)	7.66	0.006
Family Environment Scale (FES), Least Square Mean (CI)#	**Control (n = 968)**	50.1 (49.1-51.2)	55.0 (540-55.9)	51.46	<0.001
Father		**UK**	**US**	**F Value**	**Probability Value**
Coping Health Inventory for Parents (CHIP),	**Coping I: Family integration, cooperation and optimistic definition of situation (n = 637)**	31.3 (29.2-22.2)	37.1 (35.3-38.9)	21.95	<0.001

Least Square Mean (CI)#	**Coping II: Social support, self-esteem, and psychological stability (n = 637)**	19.4 (17.6-21.2)	23.6 (22.1-25.2)	15.72	<0.001
	Coping III: understanding medical situation thru communication with other parents and medical staff (n = 636)	10.2 (9.3-11.1)	12.1 (11.3-12.9)	11.73	<0.001
Family Environment Scale (FES), Least Square Mean (CI)#	**Control (n = 635)**	50.0 (48.6-51.5)	54.7 (53.5-56.0)	29.2	<0.001

Table 5: Parental scores on instruments, agreement between parents, and tests for differences

Table 5. Parental scores on instruments, agreement between parents, and tests for differences Instruments		Agreement between parents' scores	Mothers' scores	Fathers' scores	Comparison of parents' scores χ^2/F Value	Probability Value
Hospital Anxiety and Depression Scale (HADS): Anxiety, n (%)+	**Normal (0-7)**	0.35	210 (57.7)	247 (67.9)	410.7	<0.001
	Borderline or Abnormal (8-21)		154 (42.3)	117 (32.1)		
Hospital Anxiety and Depression Scale (HADS): Depression, n (%)+	**Normal (0-7)**	0.36	321 (87.0)	318 (86.9)	1.86	0.173
	Borderline or Abnormal (8-21)		45 (12.3)	48 (13.1)		
Posttraumatic Distress Scale (PTDS), n (%)+	**Mild (<=10)**	0.24	271 (82.6)	286 (87.2)	42.8	<0.001
	Moderate/Severe (11-34 plus)		57 (17.4)	42 (12.8)		
Social Readjustment Rating Scale (SRRS), n (%)+	**Little or Mild (0-199)**	0.27	561 (90.3)	568 (91.5)	0.9	0.341
	Moderate/Serious/Major (200-999)		60 (9.7)	53 (8.5)		
Parental Anxiety (STAI), n (%)+	**Low Anxiety (20-39)**	0.12	131 (66.5)	143 (72.6)	5.55	0.019
	Anxiety (40-80)		66 (33.5)	54 (27.4)		
Pediatric Inventory for	**Communication Frequency (n = 572)**	0.58	16.9 (16.3 – 17.5)	15.9 (15.3 – 16.5)	21.2	<0.001

Parents (PIP), Least Square Mean (CI)#	**Emotional Distress Frequency (n = 570)**	0.65	32.5(31.2 – 33.7)	30.3 (29.0 – 31.5)	32.5	<0.001
	Medical Care Frequency (n = 572)	0.51	13.3 (12.7 – 13.9)	13.1 (12.5 – 13.7)	1.20	0.275
	Role Function Frequency (n = 570)	0.57	17.6 (16.9-18.2)	17.0 (16.3 – 17.6)	7.56	0.006
	Total Frequency (n = 568)	0.67	80.3 (77.4 – 83.1)	76.2 (73.3 – 79.1)	19.9	<0.001
	Communication Difficulty (n = 551)	0.56	15.8 (15.1 – 16.4)	15.1 (14.4 – 15.7)	10.58	0.001
	Emotional Distress Difficulty (n = 548)	0.56	34.8 (33.3 – 36.3)	31.7 (30.2 – 33.2)	33.03	<0.001
	Medical Care Difficulty (n = 552)	0.50	12.6 (12.0 – 13.1)	12.2 (11.8 – 12.8)	2.55	0.111
	Role Function Difficulty (n = 547)	0.54	17.5 (16.8 – 18.3)	16.8 (16.0 – 17.5)	7.07	0.008
	Total Difficulty (n = 545)	0.59	80.6 (77.4 – 83.8)	75.7 (72.5 – 80.0)	20.0	<0.001
Coping Health Inventory for Parents (CHIP), Least Square Mean (CI)#	**Coping I: Family integration, cooperation and optimistic definition of situation (n = 571)**	0.50	39.8 (38.3 – 41.3)	35.1 (33.6 – 36.6)	66.0	<0.001
	Coping II: Social support, self-esteem, and psychological stability (n = 567)	0.40	26.8 (25.4 – 28.2)	22.2 (20.8 – 23.6)	61.2	<0.001

	Coping III: understanding medical situation thru communication with other parents and medical staff (n = 567)	0.40	14.1 (13.4 – 14.8)	11.5 (10.8 – 12.2)	78.3	<0.001
Family Environment Scale (FES), Least Square Mean (CI)#	**Cohesion (n= 580)**	0.50	54.2 (52.8 – 55.5)	53.6 (52.3 – 55.0)	1.11	0.292
	Expressiveness (n = 580)	0.44	52.6 (51.4 – 53.9)	49.9 (48.6 – 51.2)	28.8	<0.001
	Conflict (n = 580)	0.59	46.2 (45.0 – 47.4)	46.3 (45.0 – 47.5)	0.03	0.859
	Organization (n = 580)	0.55	53.0 (51.7 – 54.2)	51.1 (49.9 – 52.4)	17.84	<0.001
	Control (n = 580)	0.43	52.4 (51.3 – 53.5)	52.7 (51.2 – 53.4)	0.04	0.843
NOTE: + Weighted Kappa, test statistic for differences was McNemar χ^2; # Interclass Correlation Coefficients, test statistics for differences was F-Test. All models controlled for study site and heart condition; n = Number; CI = 95th Percentile Confidence Intervals.						

Supplemental Table 1. Disease Complexity by Parent Gender									
		Healthy Control Scores	Overall Sample*	Heart Condition					
				Mild CHD	BV	SV	CM	Pacemaker	ACH
Mother									
Hospital Anxiety and Depression Scale (HADS): Anxiety, n (%)[LR]	**Normal (0-7)**	61.5%[+,#]	316 (51.6)	64 (60.4)	159 (54.1)	25 (37.3)[+]	14 (37.8)[#]	31 (58.5)	22 (41.5)
	Borderline or Abnormal (8-21)	38.5%	296 (48.4)	42 (39.6)	135 (45.9)	42 (62.7)	23 (62.2)	22 (41.5)	31 (58.5)
Hospital Anxiety and Depression Scale (HADS): Depression, n (%)[LR]	**Normal (0-7)**	82.8%	508 (83)	88 (83)	254 (86.4)	50 (74.6)	28 (75.7)	46 (86.8)	41 (77.4)
	Borderline or Abnormal (8-21)	17.2%	104 (17)	18 (17)	40 (13.6)	17 (25.4)	9 (24.3)	7 (13.2)	12 (22.6)
Posttraumatic Distress Scale (PTDS), n (%)[LR]	**Mild (<=10)**		1004 (79.1)	173 (90.1)	411 (81.9)	80 (65)	39 (61.9)	67 (72)	119 (81)
	Moderate/Severe (11-34 plus)		265 (20.9)	19 (9.9)[1,2,3]	91 (18.1)[4,5]	43 (35)[1,4]	24 (38.1)[2,5]	26 (28)[3]	28 (19.1)
Social Readjustment Rating Scale (SRRS), n (%)[LR]	**Little or Mild (0-199)**		1023 (90.1)	179 (93.7)	472 (90.8)	107 (89.2)	56 (88.9)	81 (83.5)	124 (88.6)
	Moderate/Serious/Major (200-999)		112 (9.9)	12 (6.3)	48 (9.2)	13 (10.8)	7 (11.1)	16 (16.5)	16 (11.4)

Parental Anxiety (STAI), n (%)[LR]	**Low Anxiety (20-39)**		625 (63.4)	116 (70.7)	275 (61.1)	67 (60.4)	40 (69)	50 (58.1)	74 (65.5)
	Anxiety (40-80)		361 (36.6)	48 (29.3)	175 (38.9)	44 (39.6)	18 (31)	36 (41.9)	39 (34.5)
Pediatric Inventory for Parents (PIP), Least Square Mean (CI) [GLM]	**Communication Frequency (n = 966)**		16.4 (5.9)	15.3 (14.4 - 16.3)[6]	16.3 (15.8 - 16.9)[7]	19.1 (17.9 - 20.3)[6,7]	17.8 (16.2 - 19.4)	17.6 (16.4 - 18.9)	17.1 (16.1 - 18.2)
	Emotional Distress Frequency (n = 963)		31.5 (12)	28.9 (27 - 30.9)[8,9,10]	31.4 (30.2 - 32.6)[11]	37.8 (35.4 - 40.3)[8,11,12]	35.9 (32.6 - 39.3)[9]	32.4 (29.9 - 34.9)[12]	33.6 (31.4 - 35.8)[10]
	Medical Care Frequency (n = 966)		13.3 (5.7)	12.5 (11.6 - 13.5)[13]	13.3 (12.7 - 13.8)[14]	16.9 (15.7 - 18.1)[13,14,15,16,17]	13 (11.4 - 14.6)[15]	14 (12.8 - 15.2)[16]	13.4 (12.4 - 14.5)[17]
	Role Function Frequency (n = 962)		17.7 (6.5)	16.4 (15.4 - 17.5)[18]	17.8 (17.2 - 18.4)[19]	20.4 (19 - 21.8)[18,19]	17.9 (16.1 - 19.7)	18.7 (17.3 - 20.1)	18.4 (17.2 - 19.6)
	Total Frequency (n = 959)		78.8 (27.5)	72.9 (68.4 - 77.4)[20]	78.7 (76 - 81.4)[21]	94.3 (88.7 - 100)[20,21,22]	84.8 (77.1 - 92.4)	82.6 (76.9 - 88.4)	82.4 (77.4 - 87.4)22
	Communication Difficulty (n = 952)		15.1 (5.9)	14.7 (13.8 - 15.7)[23]	14.8 (14.2 - 15.4)[24]	17.3 (16 - 18.5)[23,24]	16.4 (14.7 - 18.1)	16.4 (15.2 - 17.7)	16.4 (15.3 - 17.5)
	Emotional Distress		33.3 (14.2)	31.7 (29.3 - 34.1)[25]	33.1 (31.7 - 34.5)[26]	39.2 (36.2 - 42.2)[25,26]	36.9 (32.9 - 41)	35 (32 - 38)	35.8 (33.2 - 38.4)

	Difficulty (n = 949)								
	Medical Care Difficulty (n = 952)		12.3 (5.3)	11.7 (10.8 - 12.5)[27]	12.5 (12 - 13.1)	14.2 (13.1 - 15.3)[27]	12.4 (10.9 - 13.9)	13.2 (12.1 - 14.3)	12.9 (12 - 13.9)
	Role Function Difficulty (n = 948)		17.3 (7)	16.2 (15 - 17.4)[28]	17.3 (16.6 - 18)	19.4 (17.9 - 20.9)[28]	17.4 (15.4 - 19.5)	18.9 (17.4 - 20.4)	18.5 (17.2 - 19.8)
	Total Difficulty (n = 945)		77.9 (29.9)	74 (69.1 - 79)[29]	77.7 (74.7 - 80.7)[30]	89.8 (83.5 - 96.1)[29.30]	83.3 (74.7 - 91.9)	83.5 (77.1 - 89.8)	83.2 (77.7 - 88.8)
Coping Health Inventory for Parents (CHIP), Least Square Mean (CI) [GLM]	**Coping I: Family integration, cooperation and optimistic definition of situation (n = 957)**		38.9 (12.8)	35.9 (33.7 - 38)[31,32]	40 (38.7 - 41.2)[31]	41.7 (39 - 44.4)[32,33]	40.6 (37 - 44.2)	40.5 (37.8 - 43.2)	36.3 (33.9 - 38.7)[33]
	Coping II: Social support, self-esteem, and psychological stability (n = 952)		26.8 (12.5)	25.6 (23.5 - 27.7)	27.5 (26.2 - 28.7)	29.9 (27.3 - 32.5)	28.9 (25.4 - 32.5)	27.5 (24.8 - 30.2)	24.8 (22.5 - 27.1)

	Coping III: understanding medical situation thru communication with other parents and medical staff (n = 951)		13.7 (6.3)	11.3 (10.3 - 12.4)[34,35,36,37,38]	13.9 (13.3 - 14.5)[34,39]	16.2 (14.9 - 17.5)[35,39]	15.8 (14 - 17.6)[36]	14.8 (13.5 - 16.1)[37]	14.3 (13.1 - 15.4)[38]
Family Environment Scale (FES), Least Square Mean (CI) [GLM]	**Cohesion (n = 972)**	50.0 (10.0)[$,%,^,&]	53.8 (13)	53.8 (51.6 - 56)[$]	53.9 (52.6 - 55.2)[%]	53.7 (50.9 - 56.4)	53.3 (49.6 - 57)	55.4 (52.6 - 58.2)[^]	53.8 (51.4 - 56.2)[&]
	Expressiveness (n = 971)	50.0 (10.0)[@@]	51.7 (11.5)	53.8 (51.9 - 55.7)[@@]	51.8 (50.6 - 52.9)	49.6 (47.2 - 52)	52.1 (48.9 - 55.4)	51.6 (49.2 - 54.1)	51.5 (49.4 - 53.7)
	Conflict (n = 972)	50.0 (10.0)[##]	47.4 (11.3)	47.3 (45.4 - 49.2)	47.2 (46.1 - 48.4)[##]	49.5 (47.1 - 51.9)	47.4 (44.2 - 50.6)	46.2 (43.8 - 48.6)	46.6 (44.5 - 48.7)
	Organization (n = 971)	50.0 (10.0)[$$]	52 (11)	51.3 (49.5 - 53.2)	52.7 (51.6 - 53.8)[$$]	53.1 (50.8 - 55.4)	51.5 (48.4 - 54.6)	50.8 (48.5 - 53.1)	52.7 (50.7 - 54.7)
	Control (n = 971)	50.0 (10.0)[%%,^^]	53 (10.1)	52.3 (50.7 - 54)	52.9 (51.9 - 53.9)[%%]	53 (50.9 - 55.1)	52.8 (50 - 55.5)	50.3 (48.2 - 52.4)	53.7 (51.8 - 55.5)[^^]
Father									
Hospital Anxiety and Depression Scale (HADS): Anxiety, n (%) [LR]	**Normal (0-7)**	73.6%	266 (68.4)	60 (83.3)	122 (64.6)	18 (52.9)	17 (70.8)	21 (67.7)	26 (70.3)
	Borderline or Abnormal (8-21)	26.4%	123 (31.6)	12 (16.7)[a,b]	67 (35.5)[a]	16 (47.1)[b]	7 (29.2)	10 (32.3)	11 (29.7)

Hospital Anxiety and Depression Scale (HADS): Depression, n (%)[LR]	**Normal (0-7)**	84.6%[&&]	338 (86.5)	66 (91.7)	166 (87.4)	25 (73.5)	15 (62.5)[&&]	29 (90.6)	35 (94.6)
	Borderline or Abnormal (8-21)	15.4%	53 (13.6)	6 (8.3)	24 (12.6)	9 (26.5)	9 (37.5)	3 (9.4)	2 (5.4)
Posttraumatic Distress Scale (PTDS), n (%)[LR]	**Mild (<=10)**		299 (86.9)	40 (95.2)	138 (90.8)	17 (77.3)	11 (68.8)	24 (82.8)	42 (84)
	Moderate/Severe (11-34 plus)		45 (13.1)	2 (4.8)	14 (9.2)	5 (22.7)	5 (31.3)	5 (17.2)	8 (16)
Social Readjustment Rating Scale (SRRS), n (%)[LR]	**Little or Mild (0-199)**		636 (91)	121 (97.6)	303 (92.4)	50 (84.8)	25 (83.3)	46 (82.1)	89 (89)
	Moderate/Serious/Major (200-999)		63 (9)	3 (2.4)[c,d]	25 (7.6)	9 (15.3)[c]	5 (16.7)	10 (17.9)[d]	11 (11)
Parental Anxiety (STAI), n (%)[LR]	**Low Anxiety (20-39)**		253 (72.5)	38 (73.1)	125 (77.2)	17 (56.7)	9 (50)	20 (76.9)	43 (71.7)
	Anxiety (40-80)		96 (27.5)	14 (26.9)	37 (22.8)	13 (43.3)	9 (50)	6 (23.1)	17 (28.3)
Pediatric Inventory for Parents (PIP), Least Square Mean (CI) [GLM]	**Communication Frequency (n = 638)**		15 (5.2)	14.7 (13.7 - 15.7)	14.8 (14.2 - 15.5)	17 (15.5 - 18.4)	16.7 (14.8 - 18.6)	17 (15.7 - 18.4)	14.7 (13.6 - 15.8)
	Emotional Distress Frequency (n = 636)		28.3 (10.6)	26.7 (24.5 - 28.8)	28.1 (26.7 - 29.4)	31.8 (28.8 - 34.9)	32.5 (28.6 - 36.4)	31.9 (29 - 34.7)	28.3 (26 - 30.5)

	Medical Care Frequency (n = 638)		12.6 (5.2)	12.6 (11.6 - 13.6)	12.2 (11.6 - 12.9)[e]	13.9 (12.4 - 15.4)	13.5 (11.6 - 15.4)	14.5 (13.2 - 15.9)[e]	12.2 (11.1 - 13.3)
	Role Function Frequency (n = 638)		16.4 (5.9)	15.3 (14.1 - 16.5)[f]	16.3 (15.6 - 17)	17.9 (16.2 - 19.6)	18 (15.8 - 20.1)	18.1 (16.6 - 19.7)[f]	16.1 (14.9 - 17.4)
	Total Frequency (n = 636)		72.3 (24.6)	69.3 (64.4 - 74.2)	71.4 (68.4 - 74.5)	80.6 (73.5 - 87.7)	80.7 (71.7 - 89.7)	81.3 (74.7 - 87.9)	71.4 (66.2 - 76.6)
	Communication Difficulty (n = 620)		14.1 (5.2)	13.5 (12.5 - 14.6)	14.1 (13.4 - 14.7)	16.3 (14.8 - 17.8)	14.6 (12.7 - 16.5)	15.4 (14 - 16.8)	14.4 (13.3 - 15.6)
	Emotional Distress Difficulty (n = 618)		29.6 (12.5)	27.2 (24.7 - 29.7)[g]	29.5 (27.9 - 31)	34.4 (30.8 - 38)[g]	34.2 (29.7 - 38.8)	33.1 (29.7 - 36.4)	30.1 (27.4 - 32.8)
	Medical Care Difficulty (n = 621)		11.7 (4.9)	11.6 (10.6 - 12.6)	11.8 (11.2 - 12.4)	12.7 (11.3 - 14.1)	12.1 (10.3 - 13.9)	13.2 (11.9 - 14.5)	11.8 (10.7 - 12.8)
	Role Function Difficulty (n = 619)		16 (6.5)	15 (13.7 - 16.3)	15.9 (15.1 - 16.8)	17.6 (15.7 - 19.4)	17 (14.6 - 19.4)	18 (16.2 - 19.7)	16.1 (14.7 - 17.5)
	Total Difficulty (n = 617)		71.5 (27)	67.5 (62 - 72.9)	71.3 (67.9 - 74.8)	80.9 (73.1 - 88.7)	77.9 (68.1 - 87.8)	79.4 (72.1 - 86.7)	72.5 (66.7 - 78.3)

	Coping I: Family integration, cooperation and optimistic definition of situation (n = 640)		34.7 (15.1)	30.5 (27.5 - 33.5)[h]	35.3 (33.4 - 37.1)	38.7 (34.4 - 43)[h]	31.8 (26.3 - 37.3)	34.7 (30.7 - 38.7)	34.2 (31.1 - 37.4)
	Coping II: Social support, self-esteem, and psychological stability (n = 639)		22 (13.1)	19.3 (16.7 - 21.9)	22.9 (21.3 - 24.6)	25.7 (22 - 29.5)	20.1 (15.3 - 24.8)	20.9 (17.4 - 24.3)	21.6 (18.8 - 24.3)
Coping Health Inventory for Parents (CHIP), Least Square Mean (CI) [GLM]	**Coping III: understanding medical situation thru communication with other parents and medical staff (n = 638)**		11 (6.6)	8.9 (7.6 - 10.2)[i,j]	11.2 (10.4 - 12)[i]	14.1 (12.3 - 16)[j]	11 (8.7 - 13.4)	10.6 (8.9 - 12.4)	11.5 (10.2 - 12.9)
	Cohesion (n = 644)	50.0 (10.0)[**,+++]	53.8 (11.7)	55.7 (53.4 - 58.1)[**]	54.4 (52.9 - 55.8)[++]	52.2 (48.9 - 55.5)	50.2 (45.8 - 54.5)	53.7 (50.6 - 56.8)	53.6 (51.1 - 56)
Family Environment Scale (FES),	**Expressiveness (n = 645)**	50.0 (10.0)	49.5 (11.7)	50.5 (48.2 - 52.9)	49.9 (48.4 - 51.3)	50.1 (46.8 - 53.5)	49.9 (45.5 - 54.3)	49.2 (46 - 52.3)	47.7 (45.2 - 50.2)

Least Square Mean (CI) [GLM]	**Conflict (n = 644)**	50.0 (10.0)[@@@, ###,$$$]	46.5 (10.9)	45.5 (43.3 - 47.8)[@@@]	46.4 (45 - 47.8)[###]	45.6 (42.5 - 48.8)	46.7 (42.5 - 50.8)	46.6 (43.7 - 49.5)	45.8 (43.5 - 48.2)[$$$]
	Organization (n = 644)	50.0 (10.0)	50.8 (11.6)	51.2 (48.9 - 53.5)	51.5 (50.1 - 52.9)	53 (49.7 - 56.4)	49.5 (45.2 - 53.8)	47.9 (44.8 - 50.9)	51.2 (48.8 - 53.7)
	Control (n = 644)	50.0 (10.0)	52.6 (10.7)	52.1 (49.9 - 54.2)	52.3 (51 - 53.6)	53.5 (50.5 - 56.5)	52 (48 - 55.9)	51.5 (48.7 - 54.3)	52.8 (50.6 - 55.1)

NOTE: *Overall Sample results were unweighted; scores for continuous measures were reported as means (standard deviations). Mild = Mild CHD (two-ventricle unrepaired CHD); BV = Two-ventricle with S/p heart surgery (not transplant) and/or catheter based intervention; SV = Single-ventricle S/p heart surgery (not transplant) and/or catheter based intervention; CM = Cardiomyopathy; Pacemaker = Pacemaker or ICD; ACH = Acquired heart disease. [LR]=Logistic regression was used for between group comparisons. [GLM]=General linear modeling was used to between group comparisons. n = Number. CI = 95th Percentile Confidence Intervals. χ^2=Chi Square. z = z Values. t = t Values. p = probability values

Comparisons to Healthy Controls scores:

[+]χ^2=11.95, p<0.001; [#] χ^2=7.14, p=0.007; [$] t=2.89, p=0.004; [%] t=4.02, p <0.001; [^] t=-3.42 p<0.001; [&] t=2.68, p =0.008; [@@] t=3.13 p=0.002; [##] t=2.87, p=0.004; [$$] t=2.77, p=0.006; [%%] t=2.91, p=0.003, [^^] t=2.75, p=0.007; [&&] χ^2=7.32, p=0.007; [**] t=4.12, p<0.001; [++] t=4.32, p<0.001; [@@@] t=3.37; p<0.001;[###] t=3.70, p<0.001;[$$$] t=3.24 p=0.001. Uncorrected p values are reported therefore a p value of p <0.008 (6 comparisons at a p<0.05 level) would be considered statistically significant.

Comparisons for Mothers' scores:

[1] z=4.75, p<0.001. [2] z=4.59, p<0.001. [3] z=3.55, p=0.006. [4] z=3.70, p=0.003.[5] t=3.48, p=0.008. [6] t=4.81, p<0.001. [7] t=4.11, p<0.001. [8] t=5.59, p<0.001. [9] t=6.57, p=0.006. [10] t=3.26, p=0.017. [11] t=4.71, p<0.001. [12] t=-3.05, p=0.035. [13] t=5.69, p<0.001. [14] t=5.48, p<0.001. [15] t=-3.8, p=0.002. [16] t=-3.38, p=0.011. [17] t=-4.24, p<0.001. [18] t=4.51, p<0.001. [19] t=3.43, p=0.009.

[20] t=5.83, p<0.001. [21] t=4.96, p<0.001. [22] t=-3.07, p=0.033. [23] t=3.18, p=0.023. [24] t=3.56, p=0.006. [25] t=3.89, p=0.002. [26] t=3.65, p=0.004. [27] t=3.57, p=0.006. [28] t=3.33, p=0.014.

[29] t=3.89, p=0.002. [30] t=3.46, p=0.008. [31] t=3.43, p=0.009. [32] t=3.37, p=0.012. [33] t=-2.95, p=0.049. [34] t=4.38, p<0.001. [35] t=5.71, p<0.001. [36] t=4.28, p<0.001. [37] t=4.08, p<0.001.

[38] t=3.85, p=0.002. [39] t=3.12, p=0.028. All between-group models controlled for study site. Bonferroni Correction p values are reported.

Comparisons for Fathers' scores:

[a] z=2.98, p=0.043. [b] z=3.08, p=0.031. [c] z=3.15, p=0.024. [d] z=3.15, p=0.025. [e] t=2.99, p=0.043. [f] t=2.95, p<0.05. [g] t=3.2, p=0.022. [h] t=3.08, p=0.032. [i] t=3.2, p=0.022. [j] t=4.51, p<0.001. All between-group models controlled for study site. Bonferroni Correction p values are reported.