Cardiology in the Young

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Cite this article: Bainton J, Trachtenberg F, McCrindle BW, Wang K, Boruta R, Brosig CL, Egerson DA, Sood E, Calderon J, Doman T, Golub K, Graham A, Haas K, Hamstra M, Lindauer B, Sylvester D, Woodard F, Young-Borkowski L, and Mussatto KA (2023) Prevalence and associated factors of post-traumatic stress disorder in parents whose infants have single ventricle heart disease. *Cardiology in the Young* **33**: 2171–2180. doi: 10.1017/S1047951122004012

Received: 7 September 2021
Revised: 20 October 2022
Accepted: 24 November 2022
First published online: 5 January 2023

Keywords:
Norwood; CHD; post-traumatic stress disorder; parent; stress; coping

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Prevalence and associated factors of post-traumatic stress disorder in parents whose infants have single ventricle heart disease

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Abstract

Introduction: Post-traumatic stress disorder occurs in parents of infants with CHD, contributing to psychological distress with detrimental effects on family functioning and well-being. We sought to determine the prevalence and factors associated with post-traumatic stress disorder symptoms in parents whose infants underwent staged palliation for single ventricle heart disease. *Materials and methods:* A large longitudinal multi-centre cohort study evaluated 215 mothers and fathers for symptoms of post-traumatic stress disorder at three timepoints, including post-Norwood, post-Stage II, and a final study timepoint when the child reached approximately 16 months of age, using the self-report questionnaire Impact of Event Scale – Revised. *Results:* The prevalence of probable post-traumatic stress disorder post-Norwood surgery was 50% of mothers and 39% of fathers, decreasing to 27% of mothers and 24% of fathers by final follow-up. Intrusive symptoms such as flashbacks and nightmares and hyperarousal symptoms such as poor concentration, irritability, and sudden physical symptoms of racing heart and difficulty breathing were particularly elevated in parents. Higher levels of anxiety, reduced coping, and decreased satisfaction with parenting were significantly associated with symptoms of post-traumatic stress disorder in parents. Demographic and clinical variables such as parent education, pre-natal diagnosis, medical complications, and length of hospital stay(s) were not significantly associated with symptoms of post-traumatic stress disorder. *Discussion:* Parents whose infants underwent staged palliation for single ventricle heart disease often reported symptoms of post-traumatic stress disorder. Symptoms persisted over time and routine screening might help identify parents at-risk and prompt referral to appropriate supports.

Parents of infants with CHD have an elevated risk for mental health problems including anxiety disorders, depression, and post-traumatic stress disorder particularly in the weeks and months following heart surgery.[1] For infants who undergo a staged surgical palliation for hypoplastic left heart syndrome and associated single ventricle anomalies, parents can experience a range of stressors that could be considered traumatic events. These stressors include receipt of the infant's diagnosis, complex open-heart surgeries, intensive care stays and long-term hospitalization, unexpected complications, and general uncertainty about the infant's survival and quality of life.[2,3,4,5] Prevalence over time of post-traumatic stress disorder in mothers and fathers whose children have single ventricle heart disease and undergo multiple surgeries is not well described.

Post-traumatic stress disorder may develop in response to trauma.[6] Criteria for diagnosis outlined in the Diagnostic and Statistical Manual of Mental Disorders include intrusive memories, avoidance, changes in physical and emotional reactions, and negative changes in thinking and mood.[6] Symptoms must be present for at least 30 days and can include emotional numbing,

difficulty concentrating, an altered sense of time and place, detachment from self and others, flashbacks, nightmares, difficulty sleeping, and sudden intense fear accompanied by distressing physical symptoms (e.g., racing heart, shortness of breath).[6] A formal diagnosis involves a lengthy structured interview with a trained healthcare professional, but shorter self-report screening tools such as the Impact of Event Scale – Revised are frequently used to identify individuals with probable post-traumatic stress disorder.[7]

In the Family Adaptation study, an ancillary study to the Pediatric Heart Network Single Ventricle Reconstruction Trial,[8] it was reported that parents' perception of their well-being was worse if parents experience increased symptoms of post-traumatic stress disorder.[2] Therefore, we sought to further evaluate the prevalence of symptoms of post-traumatic stress disorder in both mothers and fathers of infants with critical CHD who underwent staged surgical palliation. We also investigated potential associations of demographic, clinical, stress, and coping variables with symptoms of post-traumatic stress disorder.

Materials and methods

Design of the study

The Family Adaptation study was ancillary to the multi-institutional Pediatric Heart Network Single Ventricle Reconstruction Trial, both previously described.[2,8] The Family Adaptation study investigated parents' perceptions of family function, quality of life, and well-being after their infant underwent the Norwood procedure.[2] The study theoretical framework was based on the Resiliency Model of Family Adaptation.[9] Parents completed 13 self-reported questionnaires at three timepoints. The first assessment was post-Norwood surgery, at approximately 8 weeks of infant age. The second assessment was post-Stage II surgery, at approximately 6 months of infant age. The final follow-up assessment took place at a study visit at end of trial when the child was approximately 16 months of age.

Participants

Parents from 9 of the 15 Single Ventricle Reconstruction Trial (SVR) centres and parents from two additional centres, whose infants underwent staged surgical palliation for hypoplastic left or hypoplastic right heart syndrome, participated in the study. Both mothers and fathers of the same infant were eligible to participate. Parents not fluent in English were excluded. Parents whose infant died or required heart transplantation were excluded from participation, and parents were automatically withdrawn from the survey study if infant death or heart transplantation occurred during the study. Data obtained prior to infant death or heart transplantation were included in the analysis. Each centre's Institutional Review Board reviewed and approved the study. Parents provided informed consent to participate.

Measures

Questionnaires were organised into booklets and each participant was invited to complete one booklet per timepoint. Questionnaires took approximately 40minutes to complete. All questionnaires are described in detail in Supplementary Table S1.

The Impact of Event Scale – Revised was used to measure symptoms of post-traumatic stress disorder in parents.[8] The 22-item self-report screening questionnaire corresponds to DSM-IV symptoms of post-traumatic stress disorder in 3 subdomains as follows: intrusion (e.g., symptoms include flashbacks, nightmares, difficulty sleeping); avoidance (e.g., symptoms include emotional numbing, altered sense of time and place, detached from self and others); and hyperarousal (e.g., symptoms include reduced concentration, irritability, and sudden onset of physical symptoms like a racing heart and trouble breathing). Data for this study were collected prior to the release of the DSM-V; however, screening tools based on DSM-IV are still widely used and reported. The questionnaire's psychometric properties have been previously established.[10] Eight questions evaluated intrusion, eight evaluated avoidance, and six evaluated hyperarousal. Parents answered the questions with respect to their infant's diagnosis and ongoing clinical course. They rated each question on a 5-point Likert scale ranging from 0, "not at all" to 4, "extremely". The highest possible total score is 88, with subdomain highest scores of 32 for intrusion, 32 for avoidance, and 24 for hyperarousal. The total score provides a continuum of risk, including no risk (0–23), possible post-traumatic stress disorder (24–32), and probable post-traumatic stress disorder (> 33).[11,12,13]

Seven additional questionnaires were used to address the study aims including: Family Information Form (demographic variables); Life Stress Inventory (total burden of stressful life events); [14] State-Trait Anxiety Index (measure of transient (state) and sustained (trait) anxiety); [15] Pediatric Inventory for Parents (level of stress and appraisal of stress in parents of child with serious or chronic disease); [16] Coping Health Inventory for Parents (measures family integration and social support of parents of child with serious or chronic disease); [9] Family Inventory of Resources for Management (measures resources that impact parent self-esteem, communication, financial well-being and social support); [9] and Inventory of Parent's Experiences (measures social support network and satisfaction with parenting).[17] For stress questionnaires (2, 3, and 4), lower scores reflected lower stress and for coping questionnaires (5, 6, and 7) higher scores reflected better coping.

Statistical analysis

All analyses were conducted using SAS v9.4 (SAS Institute Inc., Cary, NC, United States of America), with p-values < 0.05 representing statistical significance. Change in post-traumatic stress disorder scores over time and comparison of mothers to fathers were assessed via repeated measures regression modelling on study timepoint (as a categorical variable) and parent (mother versus father). Multi-variable, backwards stepwise repeated measures regression was used to model the relationships between post-traumatic stress disorder symptoms and other variables of interest, separated into demographic and clinical variables and stress and coping variables. Data from both parents were included when available, specifying infant as a repeated variable within the mixed model regression. All data from the same infant over time were correlated (compound symmetric variance structure). All variables with a p-value < 0.2 in bivariable modelling were included in the initial multi-variable model, and backwards selection performed. The State-Trait Anxiety state anxiety scores were included in bivariable modelling but not included in multi-variable modelling. The State-Trait Anxiety trait anxiety scores were included in both bivariable and multi-variable modelling.

Prior to model building, independent variables were investigated for substantial associations, defined as an absolute Pearson correlation > 0.5 for continuous variables, $p < 0.05$ on Pearson

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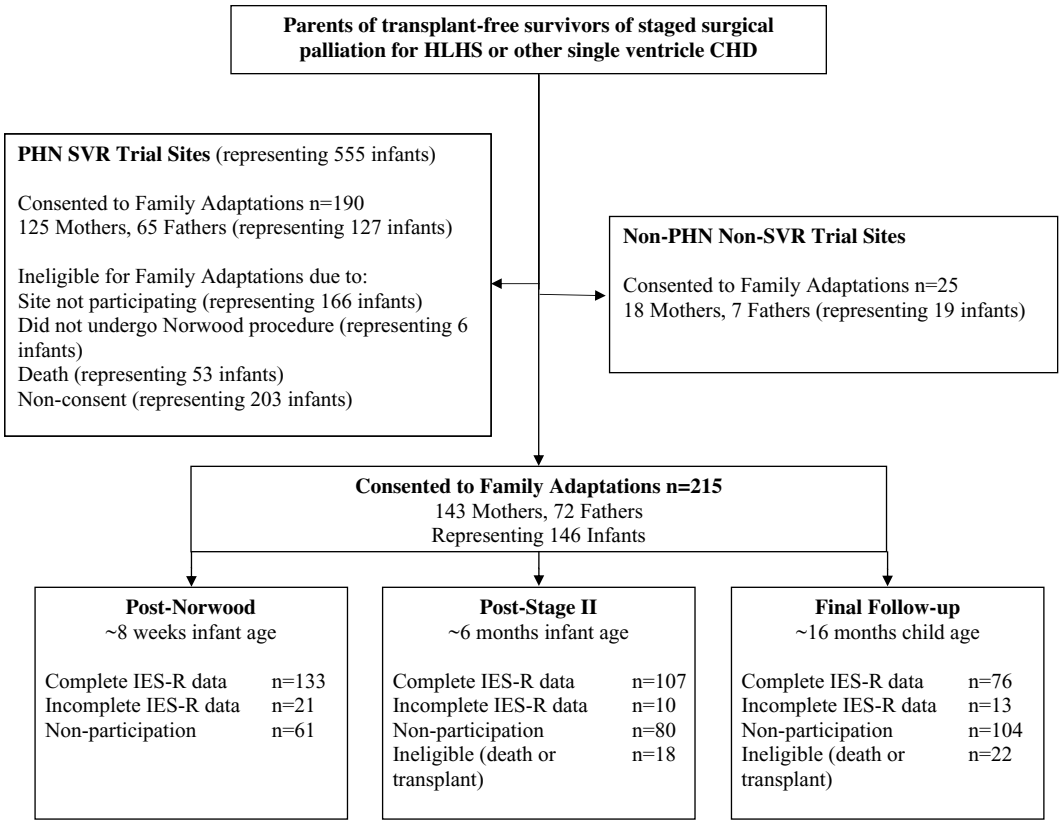

Figure 1. Consort diagram of study participant overall enrolment from trial centres and number of study participants across 3 timepoints. HLHS=hypoplastic left heart syndrome; PHN=Pediatric Heart Network; SVR=Single Ventricle Reconstruction; IES-R=Impact of Event Scale – Revised.

chi-square test for categorical variables, or $p < 0.05$ on ANOVA for the association between a continuous and a categorical variable. No substantial associations were found among the demographic and clinical variables for the total score; a small number were found for subdomains and the variable more significantly associated with outcome on bivariable modelling was chosen to proceed to multi-variable modelling, as noted in table footnotes. Some stress and coping variables had strong correlations (Supplementary Table S8), but all except trait anxiety scores were included in multi-variable modelling as these variables are designed to measure different concepts in our conceptual framework.[9] After arriving at multi-variable models for demographic/clinical variables and stress/coping variables separately, but including effects of parent and timepoint in both, all significant variables were included together in a final backwards stepwise repeated measures regression analysis to arrive at the final, combined parsimonious model. The R^2 for models (typically defined as the amount of variance in outcome explained) was calculated for these repeated measures models using the method of Jaeger et al.[18]

Results

Participant demographic information

Participants included 215 parents (143 mothers and 72 fathers) representing 146 infants (Fig 1). Demographic characteristics of parent and infants are presented in Table 1. Most infants (92%) had hypoplastic left heart syndrome, with 8% having hypoplastic right heart syndrome. Infants underwent staged surgical palliation with first surgery being the Norwood procedure in all cases. Most infants (73%) were diagnosed with CHD pre-natally. Ten percent of infants had a genetic syndrome, while 23% of infants had a non-syndromic abnormality.

Complete Impact of Event Scale – Revised data were available for 133 parents of infants post-Norwood, 107 parents post-Stage II, and 76 parents at final follow-up (Fig 1). For the participants also in SVR, timepoint 1 occurred 53 ± 113 days after Norwood surgery, timepoint 2 occurred 56 ± 129 days after Stage II surgery or before Stage II ($n = 3$), and timepoint 3 occurred 348 ± 168 days after Stage II surgery. Fourteen parents at timepoint 3 representing nine children completed assessments 2–29 days after their child's Fontan procedure. Children post-Fontan procedure were 20–32 months old versus the average age of 16 months old at final follow-up. The number of participants who became ineligible over the course of the study due to infant death or heart transplantation was 22 parents. Data obtained prior to infant death or heart transplantation were included in the analysis.

Impact of Event Scale – Revised scores over time in mothers and fathers

Total parent scores on the Impact of Event Scale – Revised were highest post-Norwood and lowest at final follow-up, for both mothers and fathers (Fig 2 ; $p < 0.001$ for time). Each of the three subdomain scores also decreased over time ($p = 0.009$ for time for intrusion, $p = 0.03$ for time for avoidance, $p < 0.001$ for time for hyperarousal). Total scores were significantly lower for fathers compared to mothers across all timepoints ($p < 0.001$ for parent; no significant interaction with time).

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Table 1. Demographic characteristics of infants, mothers, and fathers

		Post-Norwood	Post-stage II	Final follow-up
Infants at timepoint		109	80	64
Mothers completing survey*		104	78	62
Mothers with unknown information		1	0	0
Fathers completing survey*		50	39	27
Fathers with unknown information		3	1	1
Infants age at time of survey in months, mean ± sd (n)		1.7 ± 3 (103)	6.1 ± 4 (75)	15.6 ± 5 (62)
Race/Ethnicity	Caucasian/ non-Hispanic	73% (74/101)	78% (57/73)	70% (42/60)
Pre-natal diagnosis		73% (75/103)	73% (55/75)	73% (45/62)
Genetic syndrome		8% (7/83)	5% (3/65)	6% (3/51)
Non-syndromic abnormality		19% (19/98)	23% (16/71)	15% (9/59)
Mothers age at time of survey in years, mean ± sd (n)		29 ± 5 (106)	29 ± 6 (78)	31 ± 5 (63)
Mother marital status	Married or cohabitation	87% (93/107)	83% (66/80)	83% (53/64)
Mother education	High school or less	19% (21/108)	21% (17/80)	13% (8/64)
	Some college	30% (32/108)	24% (19/80)	31% (20/64)
	College	26% (28/108)	28% (22/80)	31% (20/64)
	Graduate/Professional	25% (27/108)	28% (22/80)	25% (16/64)
Fathers age at time of survey in years, mean ± sd (n)		32 ± 6 (98)	32 ± 6 (73)	34 ± 7 (61)
Father marital status	Married or cohabitation	89% (94/106)	84% (66/79)	84% (53/63)
Father education	High school or less	32% (34/105)	32% (25/79)	24% (15/63)
	Some college	30% (31/105)	28% (22/79)	25% (16/63)
	College	26% (27/105)	28% (22/79)	29% (18/63)
	Graduate/Professional	12% (13/105)	13% (10/79)	22% (14/63)

*Mothers and fathers could respond to demographic questions regarding the other parent, without the second parent being represented in the survey responses.
Mean ± sd for continuous variables and % for categorical variables. Percentages may not add up to 100% due to rounding. Parent education was a proxy for socio-economic status.

Median Impact of Event Scale – Revised total scores for mothers were 33 (IQR 21, 45) post-Norwood, 31 (IQR 18, 41) post-Stage II, and 22 (IQR 18, 41) at final follow-up, while median scores for fathers were 23 (IQR 14, 37) post-Norwood, 26 post-Stage II (IQR 11, 35), and 20 (IQR 11, 31) at final follow-up. Subdomain scores for intrusion were significantly lower for fathers than for mothers ($p < 0.001$ for parent) with a decrease over time only in mothers ($p = 0.006$ for the interaction). No difference between mothers and fathers was observed in the avoidance subdomain ($p = 0.57$ for parent), but fathers reported fewer hyperarousal symptoms compared to mothers ($p < 0.001$ for parent).

Prevalence of post-traumatic stress disorder in parents

The prevalence of probable post-traumatic stress disorder (total Impact of Event Scale – Revised > 33) was higher in mothers compared to fathers and decreased over time for both (Fig 3). The prevalence of probable post-traumatic stress disorder in mothers was 50% post-Norwood, 47% post-Stage II, and 27% at final follow-up, compared to 39%, 36%, and 24% of fathers, respectively. The percentage of mothers with possible post-traumatic stress disorder (total Impact of Event Scale – Revised score of 24–32) was 19% post-Norwood, 16% post-Stage II, and 19% at final follow-up, compared to 11%, 17% and 14% of fathers, respectively.

There were only 28 parents (20 mothers, 8 fathers) who participated at all three timepoints. Of these 28 parents, 25% had probable post-traumatic stress disorder at all three timepoints (5 mothers, 2 fathers), another 14% had possible post-traumatic stress disorder at all three timepoints (2 mothers, 2 fathers), and 29% had no risk for post-traumatic stress disorder at all three timepoints (5 mothers, 3 fathers). The sample size was too small for statistical testing and data are not reported for parents whose scores changed over time.

Factors associated with symptoms of post-traumatic stress disorder: Bivariable modelling

In the bivariable modelling, the only demographic variable significantly associated with symptoms of post-traumatic stress disorder was parent age (Table 2), with older parents reporting less symptoms of post-traumatic stress disorder ($p = 0.04$, $R^2 = 0.02$). Stress variables had much stronger associations with symptoms of post-traumatic stress disorder compared to demographic variables (Table 2, Supplementary Tables S2-S4) and had higher R^2 (up to

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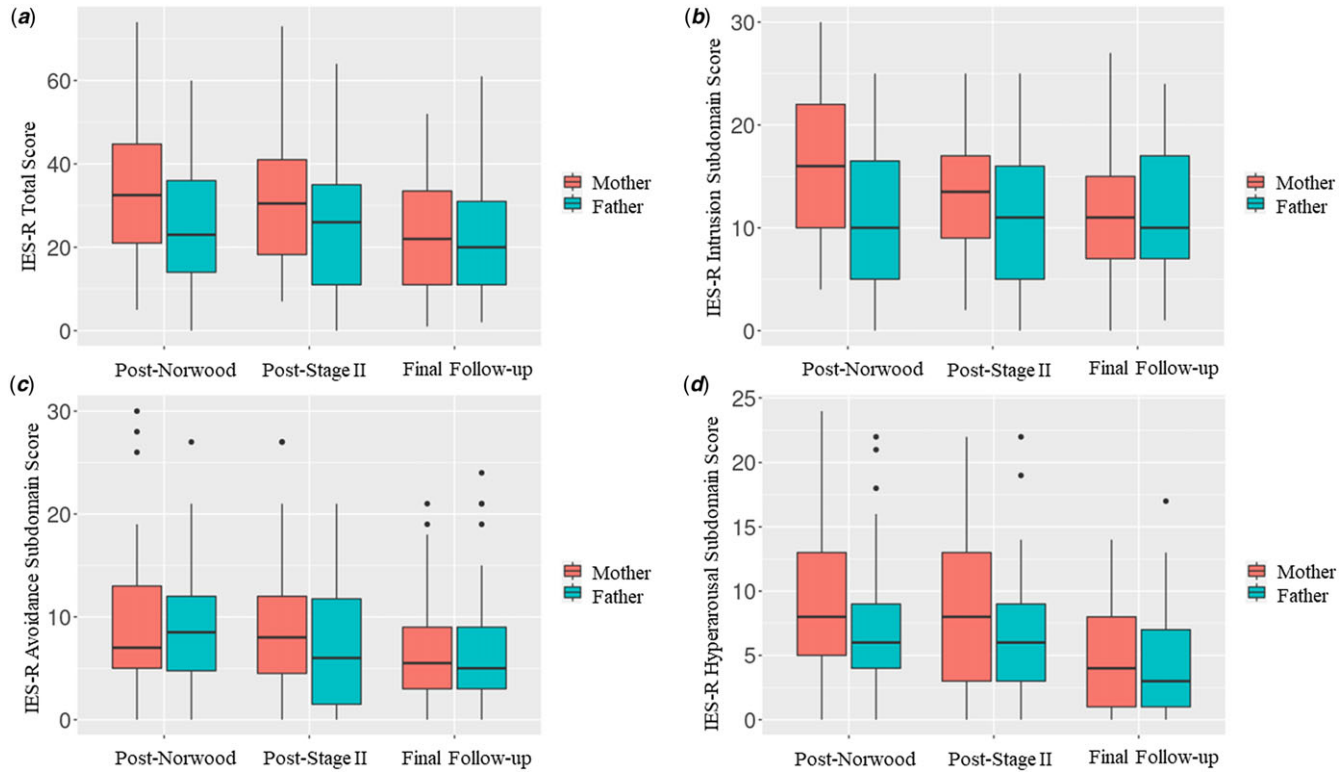

Figure 2. Median total and subdomain IES-R scores by timepoint by parent: (A) total; (B) intrusion; (C) avoidance; (D) hyperarousal. Post-Norwood ~ 8 weeks infant age. Post-Stage II ~ 6 months infant age. Final follow-up ~ 16 months child age. IES-R, Impact of Event Scale – Revised. Results of repeated measures regression demonstrate the total Impact of Event Scale – Revised scores were highest post-Norwood and lowest at final follow-up, for both mothers and fathers ($p < 0.001$ for time). Total scores were significantly lower for fathers compared to mothers across all timepoints ($p < 0.001$ for parent ; no significant interaction with time). A total Impact of Event Scale – Revised score greater than 33 indicates probable post-traumatic stress disorder. Subdomain scores also decreased over time ($p = 0.009$ for time for intrusion, $p = 0.03$ for time for avoidance, $p < 0.001$ for time for hyperarousal), though symptoms of intrusion only decreased over time for mothers ($p = 0.006$ for the interaction).

0.52 for the total Impact of Event Scale – Revised score). Higher scores of stress variables (indicating increased stress and anxiety) in the Inventory of Life Changes, State-Trait Anxiety Index, or Pediatric Inventory for Parents were significantly associated with symptoms of post-traumatic stress disorder, while lower scores of coping variables (indicating reduced coping and social support) in the Inventory of Parents Experiences or Family Inventory of Resources for Management were significantly associated with symptoms of post-traumatic stress disorder.

Factors associated with symptoms of post-traumatic stress disorder: Multi-variable modelling

In the multi-variable modelling, mothers (versus fathers) and earlier study timepoints (post-Norwood and post-Stage II) were significantly associated with more symptoms of post-traumatic stress disorder and more strongly associated with post-traumatic stress disorder than any of the demographic or clinical variables (Table 3, Supplementary Tables S5 and S7, $R^2 = 0.16–0.20$). An increased number of complications during the interstage period was the only demographic variable significantly associated with increased symptoms of avoidance ($R^2 = 0.07$) (Supplementary Table S6).

Stress variables were significantly associated with symptoms of post-traumatic stress disorder both the total and subdomain Impact of Event Scale – Revised scores. In the multi-variable modelling, higher scores in the trait anxiety score of the State-Trait Anxiety Index indicating one's tendency towards becoming anxious along with higher scores in the Pediatric Inventory for Parents indicating more frequent and more difficulty with events parents of children with serious illness experience were both significantly associated variables of symptoms of post-traumatic stress disorder (total Impact of Event Scale – Revised score, $R^2 = 0.65$). Higher scores in state anxiety scores of the State-Trait Anxiety Index indicating transient situational anxiety bring the proportion of variance to 71% (data not shown, total Impact of Event Scale – Revised score, R^2 to 0.71). Trait anxiety scores versus state anxiety scores were included in the final model because trait anxiety scores are less subject to fluctuation with change in number and degree of stressful circumstances and trait anxiety scores are strongly correlated with state anxiety scores (Supplementary Table S8). Higher scores on stress scales indicating more stress and anxiety were also significantly associated variables for increased symptoms of intrusion, avoidance, and hyperarousal (Supplementary Tables S5-S7).

Coping variables were significantly associated with symptoms of avoidance and hyperarousal. Lower scores on the Family Inventory of Resources for Management, which indicated less resources to support parent self-esteem, communication, financial well-being, and social support, were significantly associated with increased symptoms of avoidance (Supplementary Table S6). Lower scores on the Inventory of Parent Experiences, signalling less social support, and reduced satisfaction with parenting were significantly associated variables for increased symptoms of hyperarousal (Supplementary Table S7).

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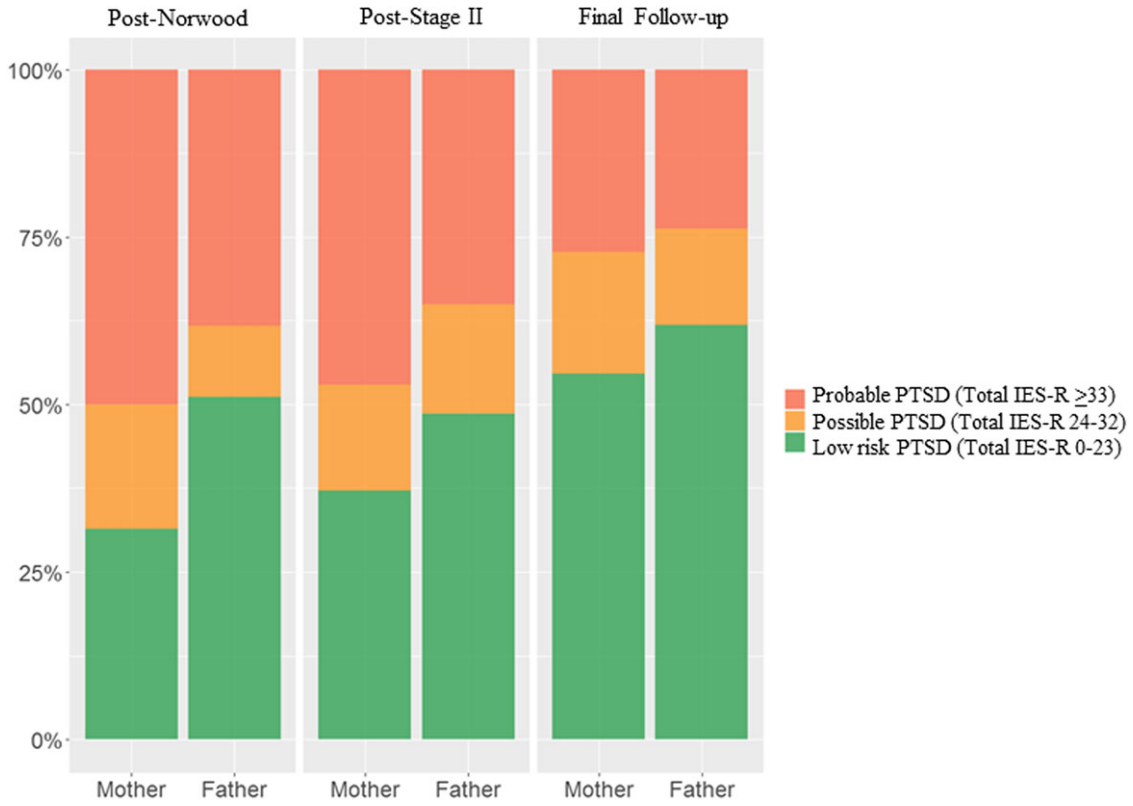

Figure 3. Stacked bar plots of IES-R scores as percentages by timepoint, stratified by parent. 50% of mothers and 39% of fathers have probable post-traumatic stress disorder post-Norwood versus 27% of mothers and 24% of fathers at the final follow-up when child is approximately 16 months old. Data presented for complete IES-R questionnaires. Missing data due to incomplete IES-R questionnaire, non-participation, and ineligibility excluded. Post-Norwood ~ 8 weeks infant age. Post-Stage II ~ 6 months infant age. Final follow-up ~ 16 months child age. PTSD=post-traumatic stress disorder; IES-R=Impact of Event Scale – Revised.

Discussion

Symptoms of post-traumatic stress disorder are prevalent and persistent in both mothers and fathers of infants with single ventricle CHD after surgical palliation. Symptoms of post-traumatic stress disorder (total Impact of Event Scale – Revised score) are significantly associated with increased parent anxiety as well as the frequency and parent's appraisal of the degree of difficulty of events they face while caring for a child with serious illness. Reduced coping and decreased satisfaction with parenting were significantly associated with two of the subdomains (avoidance and hyperarousal) of symptoms of post-traumatic stress.

In this study, as many as 50% of mothers and 39% of fathers met the criteria for probable post-traumatic stress disorder. This is considerably higher than prevalence seen in the general population, wherein the lifetime prevalence is 8% and the 12-month prevalence is 4%.[6] The disorder is associated with younger age, female gender, unemployment, single marital status, lower education, and reduced income in the general population.[6,19] The prevalence of post-traumatic stress disorder in healthy post-partum mothers, with no pre-existing mental health conditions, is 5%-13%.[20,21,22,23] Symptoms steadily decline around 6 weeks post-partum in healthy mothers.[20] Data on post-traumatic stress disorder in healthy fathers in the post-partum period are very limited although one report found no significant symptoms across the three subdomains while another reported 5% of fathers had symptoms of post-traumatic stress disorder.[24,25] Higher prevalence (19–24% of mothers and 13% of fathers) of post-traumatic stress disorder is observed in parents of preterm infants and infants with fetal abnormality.[26,27,28] The prevalence of post-traumatic stress disorder in parents of children with single ventricle CHD post-Norwood and post-Stage II palliation is comparable to that seen in bereaved mothers (42–69% of bereaved mothers).[29,30]

Of parents of children who require bypass surgery for mild to severe CHD, 25% of parents met criteria for post-traumatic stress disorder 1-month post-surgery and 15% of mothers and 10% of fathers still met criteria for post-traumatic stress disorder at 6 months post-surgery.[31,32] Data on parents further out from their child's surgery are lacking, but in a single centre study with 29 parents (16 mothers, 13 fathers) of children with hypoplastic left heart syndrome who were interviewed one time when their child was age 1–18 years old, 88% of mothers interviewed had symptoms of post-traumatic stress disorder and 66% of fathers had symptoms of post-traumatic stress disorder.[3,33,34] It is not clear if parent's symptoms of post-traumatic stress disorder fully resolve or whether post-traumatic stress disorder symptoms remain prevalent months to years after their child's surgery for single ventricle CHD. Continued research is needed.

Post-traumatic stress disorder has a high level of comorbidity with other mood disorders, anxiety disorders, disruptive behaviour disorders, chronic physical conditions, and addictions.[6,35] Poorer parent mental health including higher parent stress and anxiety has been found to be associated with poorer neurodevelopmental outcomes in children with CHD.[36] Parental post-traumatic stress symptoms severity is associated with overprotective parenting as well as emotional and behavioural problems in children who have CHD.[5] Parental post-traumatic stress is also associated with an increase in unplanned hospital visits in the first few months of their

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Table 2. Bivariable repeated measures regression modelling for IES-R total score

Variable	n	Estimate (slope)	Standard error	P-value overall	R^2
Demographic and clinical variables					
Gestational age at birth (weeks)	297	−0.85	0.85	0.322	0.01
Birth weight (kg)	301	0.00	0.00	0.818	0.00
Male gender	304	−2.60	2.43	0.286	0.01
Non-white or Hispanic race/ethnicity	296	2.14	2.77	0.440	0.01
CHD diagnosed pre-natally	304	−1.96	2.73	0.474	0.01
Genetic syndrome [a]	316	−7.15	5.30	0.207	0.02
Non-syndromic abnormality [a]	316	1.38	3.17	0.829	0.00
Age at Norwood (days)	238	0.28	0.17	0.090	0.03
Age at stage II (months)	226	0.01	0.03	0.656	0.00
LOS post-Norwood (weeks)	300	−0.02	0.04	0.570	0.00
LOS post-stage II (weeks)	284	0.04	0.03	0.305	0.01
Number of complications Norwood hospitalization	261	0.29	0.57	0.615	0.00
Number of complications interstage	179	1.20	1.49	0.424	0.01
Number of complications stage II hospitalization	246	−0.15	0.85	0.861	0.00
Number of complications post-stage II discharge	168	−0.16	1.33	0.902	0.00
Other surgery post-Norwood	278	−0.35	0.67	0.607	0.00
Other surgery post-stage II	261	1.05	1.97	0.596	0.00
Parent age (years)*	309	−0.37	0.18	0.042	0.02
Parent education	316			0.359	0.02
High school or less *versus* college degree		2.03	2.94		
Some college versus college degree		3.57	2.43		
Graduate/professional degree versus college degree		−1.00	2.60		
Single parent versus married/partner	311	6.42	3.12	0.176	0.02
Stress variables					
Inventory of Life Changes***	316	0.03	0.01	<0.0001	0.11
State-Trait Anxiety Index (State anxiety score)***	309	0.72	0.06	<0.0001	0.49
State-Trait Anxiety Index (Trait anxiety score)***	309	0.80	0.07	<0.0001	0.41
Pediatric Inventory for Parents***	239	0.31	0.03	<0.0001	0.52
Coping Variables					
Coping Health Inventory for Parents	281	−0.02	0.04	0.691	0.00
Inventory of Parents Experiences***	313	−4.63	1.08	<0.0001	0.09
Family Inventory of Resources for Management***	284	−0.23	0.04	<0.0001	0.16

*$p < 0.05$, **$p < 0.01$, ***$p < 0.001$. Total n includes a category for "unknown", which is not presented. The total n includes all participating parents across all timepoints. The statistical model used, repeated measures regression, accounts for this by treating all data entries for the same infant as correlated with each other, rather than as independent information. LOS=length of stay; IES-R=Impact of Event Scale – Revised.

infant's life.[37] Family-based psychosocial interventions that empower parents as caregivers and advocates for their children with CHD could reduce the impact of parent mental health problems and improve child outcomes.[38,39,40] Effective treatments provided early on for post-traumatic stress disorder such as cognitive-behavioural therapy have been tested in many settings showing effective results.[41] Our results demonstrate that mothers and fathers could benefit from early identification through screening for anxiety during hospitalization with continued monitoring for development of symptoms of post-traumatic stress especially symptoms that persist longer than 30 days after cardiac surgery as persistence of symptoms is part of criteria for diagnosis.

It is important to consider the reality that single ventricle CHD carries the highest morbidity of any form of CHD with a significant risk of death or heart transplantation.[42] All single ventricle CHD children require additional cardiac surgeries, catheter procedures, and a significant number will have complications including pacemaker placement, thrombotic events, stroke, seizure, protein-

https://doi.org/10.1017/S1047951122004012 Published online by Cambridge University Press

Table 3. Multi-variable repeated measures regression modelling for IES-R total score versus demographic variables and versus stress and coping variables. The final combined model was the same as the model for stress and coping variables. [a]The total n includes all participating parents across all timepoints. The statistical model used, repeated measures regression, accounts for this by treating all data entries for the same infant as correlated with each other, rather than as independent information

Variable	n	Category	Estimate (Slope)	Standard error	*p*-value overall	R^2
Demographic variables	316					0.16
Parent		Father versus Mother	−5.32	1.45	0.0005	
Timepoint		Time 2 versus Time 1	−2.16	1.61	<0.0001	
		Time 3 versus Time 1	−8.66	1.81		
Stress and coping variables	231					0.65
State-Trait Anxiety Index (Trait anxiety score)			0.49	0.07	<0.0001	
Pediatric Inventory for Parents			0.25	0.03	<0.0001	

losing enteropathy, and plastic bronchitis.[42] Complications can occur at any point in the child's life and their parents likely repeatedly experience trauma over time as their child requires further palliation that necessitates intensive care support, lengthy hospitalizations, and uncertain futures.[42] Screening for symptoms of post-traumatic stress disorder should be on-going pre-operatively through hospitalization and again post-operatively. Early identification, appropriate resource allocation to those parents who are deemed at-risk, empathic communication, and consistency in care could help support parents.[2,4,43]

Our study showed that demographic and clinical (medical or surgical) factors such as pre-natal diagnosis, complications, and length of hospital stay did not identify which parents were at greater risk to develop post-traumatic stress disorder symptoms, a finding supported by other studies.[44,45] Measures of anxiety have been associated with elevated post-traumatic stress disorder symptoms in parents of children with CHD.[45,46] Like our findings, Turgoose et al. did not find significant demographic or clinical factors that predict post-traumatic stress disorder in parents of children with CHD (any type) undergoing bypass.[45] Parents need support and education to help them adapt to the stress and challenge of raising a child with critical CHD.[47,48] Healthcare professionals are in a unique position to have a positive influence on how parents adapt to stressors by recognising symptoms in parents and referring parents to appropriate resources.

Study participant withdrawal was a limitation of this study which prevented analysis of paired parent dyads. There were too few parents with longitudinal data to address the question of change in probable post-traumatic stress disorder between timepoints per parent. The most common reason for participant withdrawal from the study was parent stress. Parents who were struggling the most may not have participated at some timepoints or may never have consented at all. It is also a possibility that survey fatigue due to the time commitment required to complete the questionnaires contributed to attrition rates. The Family Adaptation study was designed to capture the elements of a complex psychosocial process and surveys were selected based on the framework of stress appraisal, coping, and adaptation; however, all surveys contributed to the framework and allowed thorough analysis of findings. This study relied on self-report measures and the Impact of Event Scale – Revised is a screening tool versus formal diagnostic tool for post-traumatic stress disorder. The high prevalence of probable post-traumatic syndrome disorder on screening does suggest that future studies should consider a formal diagnosis through clinical standard diagnostic interview to better understand the true prevalence in this vulnerable parent population.

Attention to parent mental health and overall well-being during uncertain and stressful times is important. These results suggest more help is needed for parents, and screening and intervention might be beneficial.

Supplementary material. For supplementary material accompanying this paper visit https://doi.org/10.1017/S1047951122004012

Acknowledgements. We would like to acknowledge the NIH/NHLBI Pediatric Heart Network Family Adaptation parent participants for all data in this manuscript.

Financial support. This study was supported by grants (HL135680, HL135685, HL135683, HL135689, HL135646, HL135665, HL135678, HL135682, HL135666, HL135691, HL068270) from the NHLBI, NIH. The authors are solely responsible for the design and conduct of this study, all study analyses, the drafting and editing of the paper, and its final contents. The views expressed in this manuscript are those of the authors and do not necessarily represent the views of the National Heart, Lung, and Blood Institute; the National Institutes of Health; or the U.S. Department of Health and Human Services.

Conflicts of interest. None.

Ethical standards. The authors assert that all procedures contributing to this work comply with the ethical standards of the relevant national guidelines on human experimentation Department of Health and Human Services Code of Federal Regulations (45 CFR 46) Policy for Protection of Human Research Subjects and with the Helsinki Declaration of 1975, as revised in 2008, and have been approved by the respective institutional review boards or research ethics board of the Children's Hospital of Wisconsin, the New England Research Institutes, Nemours/Alfred I. duPont Hospital for Children, The Hospital for Sick Children, Toronto, Children's Hospital Los Angeles, Primary Children's Hospital, Children's Healthcare of Atlanta, Children's Hospital of Philadelphia, University of Michigan Health System, Boston Children's Hospital, Medical College of Wisconsin, North Carolina Consortium, Morgan Stanley Children's Hospital of New York-Presbyterian, Johns Hopkins All Children's Hospital, Medical University of South Carolina, and Cincinnati Children's Hospital and Medical Center.

References

1. Woolf-King SE, Anger A, Arnold EA, et al. Mental health among parents of children with critical congenital heart defects: a systematic review. J Am Heart Assoc 2017; 6: e004862. DOI 10.1161/JAHA.116.004862.
2. Mussatto K, Van Rompay M, Trachtenberg F, et al. Family function, quality of life and well-being in parents of infants with hypoplastic left heart

syndrome. J Fam Nurs 2021; 27: 222–234. DOI 10.1177/1074840720987309.
3. Cantwell-Bartl A, Tibballs J. Psychosocial experiences of parents of infants with HLHS in the PICU pediatr crit care med 2013.
4. Cantwell-Bartl A, Tibballs J. Psychosocial responses of parents to their infant's diagnosis hypoplastic left heart syndrome. Cardiology in Young 2014, 25:6.
5. McWhorter LG, Christofferson J, Neely T, et al. Parental post-traumatic stress, overprotective parenting, and emotional and behavioural problems for children with critical congenital heart disease. Cardiol Young 2022; 32: 738–745. DOI 10.1017/S1047951121002912. Epub 2021 Aug 9. PMID: 34365986; PMCID: PMC8825886.
6. Bromet E, Karam E, Koenen K, et al. Trauma and posttraumatic stress disorder: Global perspectives from the WHO World Mental Health Surveys. Cambridge, United Kingdom; New York, NY: Cambridge University Press, 2018.
7. Weiss DS, Marmar CR. The Impact of Event Scale-Revised. In: Wilson JP, Keane TM (eds). Assessing psychological trauma and PTSD. The Guilford Press, New York, 1997: 399–411.
8. Ohye RG, Sleeper LA, Mahony L, et al. Comparison of shunt types in the norwood procedure for Single-Ventricle lesions. N Engl J Med 2010; 362: 1980–1992. DOI 10.1056/NEJMoa0912461.
9. McCubbin HI, McCubbin MA, McCubbin HI, Thompson AI, McCubbin MA. Resiliency in Families: A Conceptual Model of Family Adjustment and Adaptation in Response to Stress and Crises, Family Measures: Stress, Coping, and Resiliency: Inventories for Research and Practice. (1st edn. Kamehameha Schools, Honolulu, HI, 2001, 1–62.
10. Beck JG, Grant DM, Read JP, et al. The impact of event scale-revised: psychometric properties in a sample of motor vehicle accident survivors. J Anxiety Disord 2008; 22: 187–198. DOI 10.1016/j.janxdis.2007.02.007.
11. Chang HP, Chen JY, Huang YH, et al. Factors associated with post-traumatic stress in mothers of preterm infants. Arch Psychiatr Nurs 2016; 30: 96–101. DOI 10.1016/j.apnu.2015.08.019.
12. Cole JCM, Moldenhauer JS, Berger K, et al. Identifying expectant parents at risk for psychological distress in response to a confirmed fetal abnormality. Arch Womens Ment Health 2016; 19: 443–453. DOI 10.1007/s00737-015-0580-6.
13. Franck L, Wray J, Gay C, et al. Predictors of parent post-traumatic stress symptoms after child hospitalization on general pediatric wards: a prospective cohort study. Int J Nurs Stud 2015; 52: 10–21. DOI 10.1016/j.ijnurstu.2014.06.011.
14. Holmes TH, Rahe RH. The social readjustment rating scale. J Psychosom Res 1967; 11: 213–218. DOI 10.1016/0022-3999(67)90010-4.
15. Spielberger CD, Gorsuch RL, Lushene R, Vagg PR, Jacobs GA. Manual for the State-Trait Anxiety Inventory. Consulting Psychologists Press, Palo Alto, CA, 1983.
16. Streisand R, Braniecki S, Tercyak KP, Kazak AE. Childhood illness-related parenting stress: the pediatric inventory for parents. J Pediatr Psychol 2001; 26: 155–162. DOI 10.1093/jpepsy/26.3.155.
17. Crnic KA. Inventory of Parent Experiences. Penn State University. Department of Psychology, University Park, PA, 1983.
18. Jaeger BC, Edwards LJ, Das K, et al. An R^2 statistic for fixed effects in the generalized linear mixed model. J Appl Stat 2017; 44: 1086–1105. DOI 10.1080/02664763.2016.1193725.
19. Ozer EJ, Best SR, Lipsey TL, et al. Predictors of posttraumatic stress disorder and symptoms in adults: a meta-analysis. Psychol Bull 2003; 129: 52–73. DOI 10.1037/0033-2909.129.1.52.
20. Dekel S, Stuebe C, Dishy G. Childbirth induced posttraumatic stress syndrome: a systematic review of prevalence and risk factors. Front Psycho 2017; 8: 1–10. DOI 10.3389/fpsyg.2017.00560.
21. De Schepper S, Vercauteren T, Tersago J, et al. Post-traumatic stress disorder after childbirth and the influence of maternity team care during labour and birth: a cohort study. Midwifery 2016; 32: 87–92. DOI 10.1016/j.midw.2015.08.010.
22. Yildiz PD, Ayers S, Phillips L. The prevalence of posttraumatic stress disorder in pregnancy and after birth: a systematic review and meta-analysis. J of Affect Disord 2017; 208: 634–645. DOI 10.1016/j.jad.2016.10.009.
23. Cook N, Ayers S, Horsch A. Maternal posttraumatic stress disorder during the perinatal period and child outcomes: a systematic review. J Affect Disord 2018; 225: 18–31. DOI 10.1016/j.jad.2017.07.045.Epub 2017-07-27.
24. Bradley R, Slade P, Leviston A. Low rates of PTSD in men attending childbirth: a preliminary study. Br J Clin Psychol 2008; 47: 295–302. DOI 10.1348/014466508X279495.
25. Ayers S, Wright DB, Wells N. Symptoms of post-traumatic stress disorder in couples after birth: association with the couple's relationship and parent-baby bond. J. Reprod. Infant Psychol 2007; 25: 40–50. DOI 10.1080/02646830601117175.
26. Aftyka A, Rybojad B, Rozalska-Walaszek I, et al. Post-traumatic stress disorder in parents of children hospitalized in the neonatal intensive care unit (NICU): medical and demographic risk factors. Psychiatr Danub 2014; 26: 347–352.
27. Aftyka A, Rybojad B, Rosa W, et al. Risk factors for the development of post-traumatic stress disorder and coping strategies in mothers and fathers following infant hospitalization in the neonatal intensive care unit. J Clin Nurs 2017; 26: 4436–4445. DOI 10.1111/jocn.13773.
28. Ionio C, Lista G, Mascheroni E, et al. Premature birth: complexities and difficulties in building the mother-child relationship. J Reprod Infant Psychol 2017; 35: 509–523. DOI 10.1080/02646838.2017.1383977.
29. Youngblut JM, Brooten D. Comparison of mothers and grandmothers physical and mental health and functioning within 6 months after child NICU/PICU death. Ital J Pediatr 2018; 44: 89. DOI 10.1186/s13052-018-0531-8.
30. Cacciatore J, Lacasse JR, Lietz CA, et al. A parent's tears: primary results from the traumatic experiences and resiliency study. Omega (Westport) 2014; 68: 183–205. DOI 10.2190/om.68.3.a.
31. Helfricht S, Latal B, Fischer JE, et al. Surgery-related posttraumatic stress disorder in parents of children undergoing cardiopulmonary bypass surgery: a prospective cohort study. Pediatr Crit Care Med 2008; 9: 217–223. DOI 10.1097/PCC.0b013e318166eec3.
32. Helfricht S, Landolt MA, Moergeli H, et al. Psychometric evaluation and validation of the german version of the acute stress disorder scale across two distinct trauma populations. J Trauma Stress 2009; 22: 476–480. DOI 10.1002/jts.20445.
33. Cantwell-Bartl AM, Tibballs J. Parenting a child at home with hypoplastic left heart syndrome: experiences of commitment, of stress, and of love. Cardiol Young 2017; 27: 1341–1348. DOI 10.1017/S1047951117000270.Epub 2017-06-06.
34. Cantwell-Bartl A. Grief and coping of parents whose child has a constant life-threatening disability, hypoplastic left heart syndrome with reference to the Dual-Process model. Death Stud 2018; 42: 569–578. DOI 10.1080/07481187.2017.1407380. Epub 2018 Apr 9.
35. Shalev A, Liberzon I, stress disorder. N Engl J Med Marmar CPost-traumatic. Post-Traumatic stress disorder. New England Journal of Medicine 2017; 376: 2459–2469. DOI 10.1056/NEJMra1612499.
36. Roberts SD, Kazazian V, Ford MK, et al. The association between parent stress, coping and mental health, and neurodevelopmental outcomes of infants with congenital heart disease. Clin Neuropsychol 2021; 35: 948–972. DOI 10.1080/13854046.2021.1896037.
37. Golfenshtein N, Hanlon AL, Lozano AJ, et al. Parental Post-traumatic stress and healthcare use in infants with complex cardiac defects. J Pediatr 2021; 238: 241–248. DOI 10.1016/j.jpeds.2021.06.073. Epub 2021 Jun 30. PMID: 34216630; PMCID: PMC8551052.
38. Gramszlo C, Karpyn A, Demianczyk AC, et al. Parent perspectives on family-Based psychosocial interventions for congenital heart disease. J Pediatr 2020; 216: 51–57.e2. DOI 10.1016/j.jpeds.2019.09.059.
39. Kasparian NA, Kan JM, Sood E, Wray J, Pincus HA, Newburger JW. Mental health care for parents of babies with congenital heart disease during intensive care unit admission: systematic review and statement of best practice. Early Hum Dev 2019; 139: 104837. DOI 10.1016/j.earlhumdev.2019.104837.Epub 2019-08-24.
40. Tesson S, Butow PN, Sholler GF, Sharpe L, Kovacs AH, Kasparian NA. Psychological interventions for people affected by childhood-onset

heart disease: a systematic review. Health Psychol 2019; 38: 151–161. DOI 10.1037/hea0000704.
41. Sijbrandij M, Olff M, Reitsma JB, et al. Treatment of acute posttraumatic stress disorder with brief cognitive behavioral therapy: a randomized controlled trial. Am J Psychiatry 2007; 164: 82–90. DOI 10.1176/ajp.2007.164.1.82.
42. Newburger JW, Sleeper LA, Gaynor JW, et al. Transplant-free survival and interventions at 6 years in the SVR trial. Circulation 2018; 137: 2246–2253. DOI 10.1161/CIRCULATIONAHA.117.029375.
43. Muscara F, McCarthy MC, Hearps SJC, et al. Featured article: trajectories of posttraumatic stress symptoms in parents of children with a serious childhood illness or injury. J Pediatr Psychol 2018; 43: 1072–1082. DOI 10.1093/jpepsy/jsy035.
44. Kolaitis G, Meentken M, Utens EMWJ. Mental health problems in parents of children with congenital heart disease. Front Pediatr 2017; 5: 1–7. DOI 10.3389/fped.2017.00102.
45. Turgoose DP, Kerr S, De Coppi P, et al. Prevalence of traumatic psychological stress reactions in children and parents following paediatric surgery: a systematic review and meta-analysis. BMJ Paediatr Open 2021; 5: e001147. DOI 10.1136/bmjpo-2021-001147. PMID: 34337164; PMCID: PMC8287603.
46. Golfenshtein N, Lisanti AJ, Cui N, Cooper BM. Predictors of Post-traumatic stress symptomology in parents of infants with congenital heart disease post-surgery and after four months. J Pediatr Nurs 2022; 62: 17–22, DOI 10.1016/j.pedn.2021.11.013. Epub 2021 Nov 25. PMID: 34839196; PMCID: PMC8942906.
47. Woolf-King SE, Arnold E, Weiss S, et al. There's no acknowledgement of what this does to people": a qualitative exploration of mental health among parents of children with critical congenital heart defects. J Clin Nurs 2018; 27: 2785–2794, DOI 10.1111/jocn.14275. Epub 2018 Mar 13. PMID: 29345005; PMCID: PMC6013337.
48. Sood E, Lisanti AJ, Woolf-King SE, et al. Parent mental health and family functioning following diagnosis of CHD: a research agenda and recommendations from the cardiac neurodevelopmental outcome collaborative. Cardiol Young 2021; 31: 900–914. DOI 10.1017/S1047951121002134. Epub 2021-06-04.