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ORIGINAL ARTICLE

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Parenting stress trajectories during infancy in infants with congenital heart disease: Comparison of single-ventricle and biventricular heart physiology

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Abstract

Objective: Parents of infants with congenital heart disease (CHD) experience increased parenting stress levels, potentially interfering with parenting practices and bear adverse family outcomes. Condition severity has been linked to parenting stress. The current study aimed to explore parenting stress trajectories over infancy in parents of infants with complex CHD, and to compare them by post-operative cardiac physiology.

Design: Data from a larger prospective cohort study was analyzed using longitudinal mixed-effects regression modeling.

Setting: Cardiac intensive care unit and outpatient clinic of a 480-bed children's hospital in the American North-Atlantic region.

Participants: Parents of infants with complex CHD (n = 90).

Measures: Parenting stress was measured via the parenting stress index-long form over four time points during infancy.

Results: Parents of infants with a single-ventricle heart experienced a decrease in total stress over time. Parents of infants with a biventricular heart experienced a decrease in attachment-related stress, and an increase in stress related to infant temperament over time. Parenting stress trajectories over time significantly differed between groups on infant temperamental subscales.

Conclusions: Findings highlight stressful and potentially risky periods for parents of infants with complex CHD, and introduce additional illness-related and psychosocial/familial aspects to the parenting stress concept. Early intervention may promote parental adaptive coping and productive parenting practices in this population.

KEYWORDS
congenital heart disease, infancy, longitudinal design, parenting stress, uni-ventricular heart

1 | INTRODUCTION

Congenital heart disease (CHD) is the most prevalent group of congenital anomalies diagnosed in approximately one percent of live births.[1] Infants with complex defects often require multiple stages of palliative and corrective surgeries early in life, followed by long hospitalizations in the cardiac intensive care unit (CICU). Infants with single-ventricle (SV) post-operative physiology (eg, Hypoplastic Left Heart Syndrome) are extremely medically fragile and require close medical attention post-discharge. They often display feeding difficulties, growth delays, and remain at risk for congestive heart failure.[2] Their health issues cause profound stress to families especially in early infancy, in which parents ought to adjust to the intensive care environment, and to the post-operative caretaking demands at home.[3] With the increasing CHD survival rates,[4] the stress is often long-lasting and has long-term implications on quality of life.[5]

Studies reported increased parenting stress in populations with CHD, compared to the general population.[3,6,7] Parenting stress is a distinct form of psychological distress experienced by parents while trying to meet the parenting role demands.[8] It has been associated with various adverse family outcomes, including poor familial quality of life and well-being, anxiety and depression among both children and parents.[9,10] It has also been predictive of poor social competence and maladaptive behaviors among children.[11,12]

The currently identified sources of parenting stress in the CHD pediatric population mostly align well with Abidin's Parenting Stress Model.[13] The model identifies certain child characteristics, parental factors, and life events outside the parent-child system, as stress-evoking, and categorizes them into three domains (child domain, parent domain, and life stress domain). Studies have shown that parenting stress in the CHD population was dominantly related to temperamental and behavioral characteristics of the children. Specifically parents reported on increased irritability, moodiness, demandingness, and feeding problems among their children, causing them stress.[7,14,15] Parental depression, anxiety, feelings of incompetence, marital problems, and low socioeconomic status were all linked to parenting stress in the CHD population.[14,16-18]

Additional illness-related factors have been identified in the literature as important stressors in the CHD population, but have yet been included in Abidin's model. Such factors relate to the intensive care environment, the illness severity, and the increased caretaking burden at home.[3,5] Only handful of studies compared parenting stress levels by the condition complexity. For instance, Torowicz et al[15] found higher stress levels in SV infants compared to biventricular (BV) physiology infants, and healthy controls at 3 months of age. Furthermore, the trajectory of stress has yet been studied, due to the paucity of longitudinal assessments.[7] The current study aimed to examine parenting stress trajectories in parents of infants with CHD over infancy, and to compare them by the condition severity (ie, post-operational cardiac physiology). Longitudinally assessing the trajectories of parenting stress over this critical period of infancy may expand our understanding regarding parental illness adjustment following the sensitive post-diagnostic/surgical period.

2 | METHODS

2.1 | Study design

The study employed a secondary analysis of data from a larger prospective cohort study, in which infant and parent outcomes were examined during five time points over infants' first year of life.

2.2 | Setting and participants

A convenience sample of infants with CHD and their parents (N = 241) was recruited from the CICU of a 480-bed children's hospital in the American North-Atlantic region. Infants were included in the sample if they underwent corrective or palliative surgery for their heart defect within their first 6 weeks of life, born >35 gestational weeks, and weighed >2000 g. Infants with other congenital anomalies or genetic syndromes (except 22q deletion and DiGeorge syndrome) were excluded from the study.

2.3 | Study procedures and data collection

The original and the current study were approved by the institutional review board. Informed consent was signed by parents. Data were obtained at hospital discharge, and during outpatient visits at 3, 6, 9, and 12 months of age. Parents filled in self-reporting questionnaires including demographic information, and parenting stress; clinical information was obtained from the medical records.

2.4 | Study variables and instruments

2.4.1 | Parenting stress index-long form

Parenting stress as the study's outcome was assessed at 3-, 6-, 9-, and 12-month visits. The parenting stress index (PSI) is a validated, standardized, self-reporting questionnaire designated for parents, measuring stressors on the domains identified in Abidin's model. The long form consists of 120 items, yielding scores over 17 subscales. Forty-seven 5-point Likert scale items measure stress over the six child domain subscales. An item for example: "My child seems to cry or fuss more often than most children." Fifty-four 5-point Likert scale items measure the seven parent domain subscales. An item for example: "I often feel guilty about the way I feel towards my child." Scores from the parent and child domains are summed to an overall score, constructing the total stress subscale. Alpha reliability coefficients for the different subscales range between .70 and .90.[13] The additional life stress domain lists 19 stressful life events (yes/no response), potentially experienced by parents outside the parent-child system (eg, divorce, troubles at work). Individual interpretation of subscales also allows to analyze specific aspects of the parent-child system.[13] All PSI scores are analyzed on a continuum, where higher scores are indicative of higher parenting stress levels.

2.4.2 | Post-operative cardiac physiology

Infants were categorized as SV or BV post-operative cardiac physiology. Cardiac functionality was assessed by a cardiologist based on post-operative echocardiograms, in accordance with the established standards.[19]

2.5 | Covariates considered for analysis

Feeding mode. Early feeding issues in CHD infants are common and are associated with other illness parameters (brain dysfunction, psychomotor issues, energy imbalance, etc). They often correlate with later neurological impairments and developmental delays diagnosed over time, therefore might confound our relationship of interest.[2,20,21] Infants were classified by their enteral feeding modes at the time of hospital discharge as exclusively orally fed (breast or bottle), or as device-assisted feeding (nasogastric tube only or oral+ tube feeding).

2.5.1 | Infant anthropometrics

Stunted growth might be a confounding issue for infants with CHD.[21] Infant weight, length, and head circumference were obtained at all visits and converted to standardized *Z*-scores, per the World Health Organization's recommendations.[22]

2.5.2 | Demographic characteristics

Demographics were collected from the medical records and via parents' self-reporting, and included infant gestational age, gender, race, ethnicity, parental education, and whether the CHD was prenatally diagnosed.

3 | DATA ANALYSES

Descriptive statistics were generated to characterize all demographic and clinical variables. Means, standard deviations, medians, and ranges were used to describe continuous variables. Frequencies and percentages were used to describe categorical variables. Fisher's exact tests were used to examine differences in demographic and clinical variables for SV vs BV infants; two-sample *t*-tests were used to compare continuous variables across the two groups. Next, separate linear mixed-effects regression models[23] for the PSI subscales were generated for each group (SV and BV) to evaluate the group's individual stress trajectory over time. The evaluation of differences in the stress trajectories between groups relied on the group × time interaction term (time as a continuous measure from 3 to 12 months). Covariates considered for the analysis were examined based on significance level .2 in bivariate and two-way covariate × time interaction models, and further by backward deletion process in the .2 significance level. Final covariates for the multivariable analysis included infant birth weight, length *Z*-scores, and feeding mode at discharge. Given the fixed sample size due to the secondary nature of the study, multiplicity was not accounted for, and statistical significance of results was interpreted in the context of clinical meaningfulness. All analyses were conducted using STATA Version 14 (xtmixed procedure).[24]

4 | RESULTS

Table 1 displays the sample's comparison of demographic and clinical characteristics by physiology group. The final sample included 90 mothers of infants with complex CHD, of whom 45 (50%) had SV post-op physiology. Mothers were mostly non-Hispanic (n = 60; 67%) white (n = 82; 91%). Infants with SV physiology, on average, had a significantly longer post-operation CICU stay than BV infants (Mean = 31.35 vs Mean = 15.84; P = .0026), and more SV infants required device-assisted feeding at the time of discharge (53% vs 20%; P = .001). Table 2 displays the group comparisons of the baseline parenting stress subscale scores at 3 months of age. Parenting stress at 3 months did not significantly differ between groups, and total stress means corresponded to the 50th percentile on the PSI.[13]

Tables 3 and 4 present results from mixed-effects regression analyses, in which PSI subscales were separately regressed over infant visits at 3, 6, 9, and 12 months. Models were adjusted for birth weight, feeding mode at discharge, and infant length *Z*-scores. Table 2 displays the groups' individual stress trajectories over time. Findings indicate a significant decrease in parenting stress in the SV group on the mood (P = .026), attachment (P = .004), role restriction (P = .043), parent domain, (P = .043), and total stress subscales (P = .031). The BV group demonstrated significant stress increase over time on the distractibility subscale (P = .003), and stress decrease on the attachment subscale (P = .002).

Table 3 displays group differences in PSI changes over time represented by group × time interaction terms. Parents of SV and BV infants significantly differed in their parenting stress trajectories over time on the distractibility (P = .002), mood (P = .009), and the child domain (P = .023) subscales. Group comparisons of the stress trajectories over time are based on model estimates. Figure 1 graphically presents the child domain stress trajectories over time, on which the SV group demonstrated a decrease in stress over time, and the BV group demonstrated an increase in stress over time.

5 | DISCUSSION

The current study aimed to explore stress trajectories in parents of infants with CHD over the first year of life, and compare them by infants' post-operational cardiac physiology.

Findings indicate that the total stress of parents in the SV group decreased over the first year of infant's life, specifically the stress resulting from attachment issues, parental role restriction, and infant's temperamental characteristics. Whereas parents of BV infants similarly demonstrated attachment-related stress decrease over time, they also demonstrated increase in stress that was related to their infant's distractibility. Significant differences were found between the two groups in stress trajectories related to infant's temperamental characteristics (ie, mood and distractibility).

Parenting stress is expected to decrease over infancy and toddlerhood in the general healthy population.[13] Studies examining stress changes in non-healthy pediatric populations such as children with ASD, cancer, and children with disabilities, presented mixed results. Stress decrease in these studies was rationalized by the parental adjustment to the situation and/or by the reduction of treatments with time.[25,26] In the CHD population, the post-operational period and the lengthy CICU stay have been described as peak stressful periods for parents.[27,28] The post-discharge period is critical as well,

TABLE 1 Demographic characteristics and growth parameter comparisons of the study sample, *N* = 90

	SV *N* = 45			BV *N* = 45			
	N (%)			*N* (%)			*P*-value[a]
Infant gender							.652
Male	32 (71)			29 (64)			
Female	13 (29)			16 (36)			
Ethnicity							.709
Hispanic	5 (11)			3 (7)			
Non-hispanic	29 (64)			31 (69)			
Unreported	11 (24)			11 (24)			
Race							.066
White	38 (84)			44 (98)			
Black	5 (11)			1 (2)			
Other	1 (2)			0 (0)			
Unreported	1 (2)			0 (0)			
Mother's education							.628
High school	2 (4)			2 (4)			
Collage	14 (31)			16 (36)			
Post-graduate degree	3 (7)			7 (16)			
Unreported	26 (58)			20 (44)			
Feeding mode at discharge							.001
Oral feeding	13 (29)			26 (58)			
Tube assisted	24 (53)			9 (20)			
Missing	8 (18)			10 (22)			
	N	Mean (SD[b])	Median (IQR[c])	*N*	Mean (SD)	Median (IQR)	
Birth weight, g	45	3310 (506)	3310 (765)	45	3397 (518)	3430 (700)	.426
Gestational age, weeks	45	38.8 (1.50)	39 (2)	43	38.9 (1.17)	39 (2)	.599
Weight at 3 months, *Z*-score[d]	27	−1.62 (1.36)	−1.62 (2.23)	32	−.89 (1.19)	−.505 (1.49)	.033
Length at 3 months, *Z*-score	25	−1.31 (1.49)	−1.17 (1.83)	32	−.47 (1.15)	−.26 (1.6)	.020
Head circumference at 3 months, *Z*-score	23	−1.39 (1.31)	−1.48 (1.67)	32	−.17 (1.02)	−.325 (1.215)	.000
Days of hospital stay	45	31.35 (31.33)	18 (22)	44	15.84 (11.03)	13 (9)	.003

[a]Group comparisons via *t*-tests for continues parameters and Fisher's exact test for categorical parameters.
[b]Standard deviation.
[c]Interquartile range.
[d]WHO growth *Z*-scores.

especially for SV infants, who remain within the inter-stage mortality danger zone following the bidirectional Glenn procedure until 4 to 6 months of age.[29] During this period, parents are overwhelmed by their child's condition, medications, and feeding problems.[27] Over time most infants stabilize, require fewer medical interventions and treatments, and feed better. Parents also learn to cope more efficiently with stress with time, and adjust to the condition.[25,30] Gaskin et al.[31] showed decrease in signs of PTSD in parents of infants who underwent cardiac surgery, as their confidence increased.

Both groups demonstrated stress decrease on the attachment subscale. Attachment related stress is often reflected in weak parent-child relationships.[13] Insecure attachment and weak infant-mother relationships have been reported in other studies of the CHD population and other chronic pediatric populations.[32,33] It is assumed that bonding and attachment issues in the CHD population stem from psychological and physical barriers due to the long hospitalizations in the CICU environment, and/or the uncertainty in the infant's survival.[34,35] Infants with complex CHD also tend to quickly lose attention and withdraw during interactions, challenging their care providers to maintain communication.[36] This, however, improves with time and may explain the stress decrease on the attachment subscale.

Further findings demonstrate differences in stress trajectories between groups on child's temperamental subscales. These findings may be attributed to temperamental changes in the pediatric CHD population evident in the literature.[37] Infants with more complex CHD conditions often demonstrate irritability and moodiness early in life,

TABLE 2 Baseline parenting stress comparisons of the study sample at 3 months of age

	SV			BV			
PSI subscales	*N*	Mean (SD)	Median (IQR)	*N*	Mean (SD)	Median (IQR)	*P*-value[a]
Child domain	32	99.37 (24.68)	97 (23.5)	34	91.88 (17.29)	90 (18)	.156
Distractibility	32	24.09 (4.89)	24 (4.5)	34	22.15 (4.46)	22 (4)	.096
Adaptability	32	25.84 (6.59)	26 (7.5)	34	24.15 (5.40)	25 (4)	.256
Reinforces Parents	31	8.16 (3.33)	7 (4)	34	8 (2.07)	7.50 (4)	.814
Demandingness	32	19.56 (5.99)	19 (7)	34	17.71 (5.23)	16 (7)	.184
Mood	32	9.88 (3.49)	10 (4.5)	34	9.29 (2.58)	9 (4)	.443
Acceptability	32	12.09 (4.69)	12 (6)	34	10.59 (3.42)	11 (4)	.139
Parent domain	32	114.21 (23.44)	115 (31)	34	110.73 (18.06)	109 (19)	.499
Competence	32	25.03 (6.13)	25 (9)	34	23.35 (4.87)	24 (4)	.222
Isolation	32	12.62 (4.55)	12 (6)	34	12.47 (3.23)	13 (3)	.874
Attachment	32	11.43 (3.26)	11 (4.5)	34	10.82 (1.81)	11 (3)	.345
Parental health	32	12.25 (2.78)	12 (2.5)	34	12.29 (3.21)	12.5 (4)	.953
Role restriction	32	17.59 (5.50)	17.5 (7)	34	17.94 (4.74)	16 (5)	.784
Depression	32	17.75 (4.55)	16 (6.5)	34	17.79 (3.82)	17.5 (3)	.966
Spouse	32	17.53 (5.29)	18 (7)	34	16.05 (4.74)	16 (4)	.238
Life stress	32	11.09 (7.91)	8 (8.5)	34	8.29 (6.73)	7 (9)	.126
Total stress	32	213.59 (43.58)	212 (46)	34	202.61 (31.71)	200.5 (32)	.244

Abbreviations: BV, bi-ventricle group; IQR, interquartile range; PSI, parenting stress index; SV, single-ventricle group; SD, standard deviation.
[a]Group comparisons via *t*-tests.

TABLE 3 Mixed-effects model results[a] for PSI subscales regressed on time[b]

Single-ventricle physiology						Bi-ventricle physiology				
PSI subscale[c]	β^b	SE[d]	95% CI[e]	*P*	*N*	β^b	SE[d]	95% CI[e]	*P*	*N*
Distractibility	−.26	0.39	(−0.98, 0.46)	.476	34	1.13	0.38	(0.37, 1.88)	.003	35
Adaptability	−.56	0.45	(−1.43, 0.33)	.221	34	.28	0.33	(−0.37, 0.93)	.403	35
Reinforces parents	−.17	0.21	(−0.57, 0.24)	.412	34	−.28	0.15	(−0.57, 0.01)	.061	35
Demandingness	−.45	0.44	(−1.31, 0.42)	.310	34	.01	0.32	(−0.61, 0.63)	.978	35
Mood	−.59	0.26	(−1.11, −0.07)	.026	34	.02	0.14	(−0.27, 0.30)	.908	35
Acceptability	−.16	0.35	(−0.86, 0.53)	.643	34	−.03	0.23	(−0.48, 0.41)	.885	35
Child domain	−2.00	1.52	(−4.98, 0.98)	.188	34	1.17	1.05	(−0.89, 3.24)	.265	35
Competence	−.54	0.41	(−1.35, 0.26)	.186	34	−.23	0.30	(−0.81, 0.36)	.451	35
Isolation	−.10	0.26	(−0.61, 0.40)	.684	34	−.33	0.24	(−0.80, 0.13)	.161	35
Attachment	−.61	0.21	(−1.02, −0.19	.004	34	−.47	0.15	(−0.77, −0.17)	.002	35
Health	−.08	0.22	(−0.51, 0.35)	.711	34	−.31	0.24	(−0.79, 0.17)	.208	35
Role restriction	−.61	0.31	(−1.21, −0.02)	.043	34	−.41	0.31	(−1.02, 0.19)	.181	35
Depression	−.36	0.36	(−1.09, 0.34)	.303	34	−.34	0.30	(−0.92, 0.25)	.258	35
Spouse	−.35	0.32	(−0.99, 0.28)	.274	34	−.31	0.31	(−0.91, 0.30)	.321	35
Parent domain	−2.53	1.25	(−4.98, −0.08)	.043	34	−2.23	1.29	(−4.76, 0.30)	.083	35
Total stress	−4.51	2.09	(−8.61, −0.41)	.031	34	−1.07	1.86	(−4.72, 2.59)	.567	35
Life stress	−.40	0.50	(−1.38, 0.58)	.421	34	.47	0.40	(−0.31, 1.25)	.240	35

[a]All models are adjusted for infant length *Z*-scores, birth weight, and feeding mode at discharge (exclusively oral feeding vs device assisted feeding).
[b]Estimates in table correspond to main effect of "Time"; Time represents the continuous independent variable.
[c]Parenting stress index subscale scores as the outcome of interest, each represents a separate multivariate model within each group, single-ventricle and bi-ventricle physiology.
[d]Standard error.
[e]95% confidence intervals.

TABLE 4 Final mixed-effects model results[a] for PSI subscales regressed on time[b], post-op cardiac physiology, and time × post-op cardiac physiology terms. $N = 69$

PSI subscale[d]	β	SE[e]	95% CI[f]	P
Distractibility				
Time	−.38	0.38	(−1.13, 0.37)	.320
BV physiology[c]	−6.37	2.05	(−10.39, −2.35)	.002
Group × time	1.63	0.51	(0.62, 2.63)	.002
Intercept	27.92			
Adaptability				
Time	−.57	0.39	(−1.34, 0.20)	.145
BV infants	−2.85	2.37	(−7.49, 1.80)	.230
Group × time	.88	0.52	(−0.14, 1.90)	.090
Intercept	24.55			
Reinforces parents				
Time	−.27	0.18	(−0.62, 0.08)	.126
BV infants	−.16	1.06	(−2.23, 1.91)	.878
Group × time	.06	0.23	(−0.40, 0.51)	.806
Intercept	11.71			
Demandingness				
Time	−.46	0.38	(−1.21, 0.29)	.227
BV infants	−1.74	2.27	(−6.19, 2.71)	.443
Group × time	.54	0.51	(−0.46, 1.55)	.290
Intercept	15.23			
Mood				
Time	−.64	0.20	(−1.04, −0.24)	.002
BV infants	−2.08	1.23	(−4.50, 0.34)	.092
Group × time	.71	0.27	(0.17, 1.24)	.009
Intercept	12.81			
Acceptability				
Time	−.22	0.29	(−0.79, 0.35)	.450
BV infants	−1.63	1.62	(−4.80, 1.54)	.313
Group × time	.26	0.39	(−0.51, 1.03)	.511
Intercept	13.67			
Child domain				
Time	−2.34	1.30	(−4.89, 0.20)	.071
BV infants	−14.16	8.15	(−30.14, 1.81)	.082
Group × time	3.94	1.73	(0.54, 7.34)	.023
Intercept	102.41			
Competence				
Time	−.54	0.33	(−1.17, 0.10)	.100
BV infants	−2.21	1.90	(−5.93, 1.52)	.245
Group × time	.41	0.43	(−0.44, 1.25)	.346
Intercept	32.00			
Isolation				
Time	−.16	0.24	(−0.64, 0.32)	.507
BV infants	−.19	1.39	(−2.90, 2.53)	.893
Group × time	−.08	0.32	(−0.72, 0.55)	.794

(Continues)

TABLE 4 (Continued)

PSI subscale[d]	β	SE[e]	95% CI[f]	P
Intercept	13.39			
Attachment				
Time	−.60	0.18	(−0.95, −0.24)	.001
BV infants	−.67	1.07	(−2.78, 1.43)	.532
Group × time	.13	0.24	(−0.34, 0.59)	.589
Intercept	14.99			
Health				
Time	−.18	0.23	(−0.64, 0.27)	.425
BV infants	.36	1.34	(−2.27, 2.99)	.788
Group × time	−.06	0.31	(−0.66, 0.54)	.842
Intercept	11.61			
Role restriction				
Time	−.57	0.30	(−1.15, 0.01)	.054
BV infants	−.37	1.72	(−3.74, 2.99)	.828
Group × time	.22	0.39	(−0.54, 0.98)	.571
Intercept	19.61			
Depression				
Time	−.47	0.33	(−1.12, 0.19)	.162
BV infants	−.90	1.69	(−4.21, 2.42)	.596
Group × time	.25	0.44	(−0.62, 1.12)	.569
Intercept	16.78			
Spouse				
Time	−.31	0.32	(−0.94, 0.32)	.340
BV infants	−.93	1.74	(−4.35, 2.49)	.593
Group × time	.06	0.43	(−0.78, 0.90)	.885
Intercept	21.65			
Parent domain				
Time	−2.78	1.23	(−5.18, −0.38)	.023
BV infants	−5.09	6.63	(−18.09, 7.90)	.442
Group × time	.84	1.62	(−2.34, 4.02)	.604
Intercept	135.73			
Total stress				
Time	−5.07	2.01	(−9.01, −1.12)	.012
BV infants	−19.75	12.31	(−43.88, 4.37)	.109
Group × time	4.80	2.65	(−0.40, 10.00)	.071
Intercept	236.24			
Life stress				
Time	−.38	0.45	(−1.25, 0.49)	.392
BV infants	−6.49	2.68	(−11.75, −1.24)	.015
Group × time	.93	0.58	(−0.21, 2.08)	.109
Intercept	1.96			

[a]All models are adjusted for infant length *Z*-scores, birth weight, and feeding mode at discharge (exclusively oral feeding vs device assisted feeding).
[b]Time as the continuous independent variable.
[c]Bi-ventricle versus single-ventricle post-op cardiac physiology.
[d]Parenting stress index subscale scores as the outcome of interest, each represents a separate multivariable model.
[e]Standard error.
[f]95% confidence intervals.

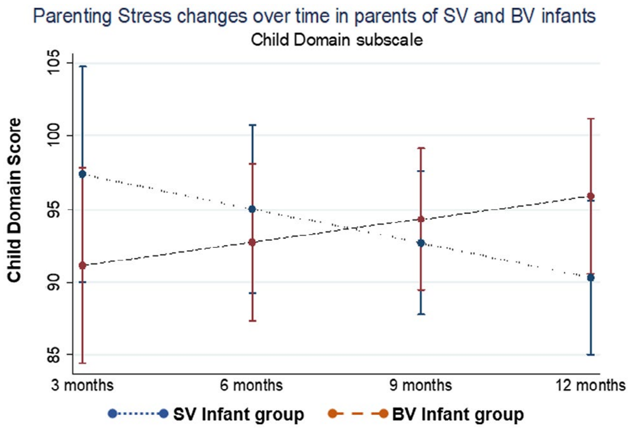

FIGURE 1 Parenting stress trajectories in parents of SV and BV infants from 3-12 months, on the child domain

which have been attributed to neurologic deficits and clinical parameters,[37] and have been reported to cause a great amount of stress to parents early in infancy.[5] With time, many infants overcome these difficulties, which and may explain the decrease in stress in the SV group. Parents of BV infants, however, demonstrate increase in stress over time on the distractibility subscale. Uzark and Jones showed via cross-sectional associations greater parental stress with age, in children with CHD between 2 and 12 years. This was explained by the increasing challenge disciplining, and setting limits to children as they age. Similarly, BV infants may follow the typical developmental route, in which infants become more distractible after the newborn stage,[38] which may explain the increase in stress on this subscale.

6 | IMPLICATIONS FOR RESEARCH, PRACTICE, AND POLICY

The longitudinal design of the current study allowed tracking parenting stress trajectories during infancy in a growing chronic pediatric population, and comparing stress trajectories by condition severity. The general decrease in stress in parents of SV infants, who are more critically ill, strengthens previous research, which highlighted the post-operational and hospital discharge as peak stressful periods. These periods are usually followed by a process of parental adjustment to the new situation of self-care at home.[5] Nevertheless, findings from the current study indicate differences in stress trajectories by illness severity, which suggest that each group has unique experiences, needs, and coping mechanisms. Golfenshtein and colleagues[39] found that early coping of mothers of infants with complex CHD included passive mechanisms, which have been previously associated with adverse outcomes, and delayed illness adjustment. Early interventions aiming to empower parents to use active coping strategies may help them to adjust to the new reality in the CICU and at home, and promote productive parenting practices.[40,41] Future research may also benefit from investigation of the different long-term coping mechanisms parents use over infancy and childhood, and characterize them by the condition severity.

While findings further show that attachment-related stress decreased over time in our sample, early attachment issues may bear long-lasting consequences on the parental practices and infant development.[42,43] Parents described difficulties in closely connecting with their infants during the CICU hospitalization.[5] A system of family care should be established for families, providing comprehensive familial support while in the CICU and beyond, and prioritizing the promotion of healthy parent-infant relationships.

7 | STUDY LIMITATIONS AND DIRECTIONS FOR FUTURE RESEARCH

The sample was recruited from a single institute, and included mainly white, non-Hispanic mothers, a fact that may limit the generalizability of results. Future research should aim for a diverse sample, including fathers as well. Studies examining both parents showed differences in parenting stress patterns.[3,14] Inclusion of other familial or maternal parameters such as family management, maternal psychosocial factors,[44,45] that account not only for parental caregiving in the hospital but also demand caretaking at home in future studies, may enhance comprehensive understanding of the phenomenon.

Although the PSI was validated in the CHD population, its ability to capture certain illness-specific aspects of the phenomenon is limited.[40] The sample of the current study demonstrated lower-than-expected stress levels, which may be attributed to parental defensive response,[13] or to the measure's inability to fully capture aspects of the phenomenon. More illness-specific parenting stress measures may provide information beyond the obtained from the general measure.[46] Furthermore, data collection has been performed almost a decade ago. Although changes have not been applied to the PSI since then, the ongoing advances in technology, surgery, medicine, and nursing care may impact the current stress experience of parents in similar situations.

Missing data and dropout were accounted for in the mix-modeling approach, and by comparisons to an imputed dataset.[47] This, however, limited the power in the study, and limited our ability to adjust for multiplicity issues (eg, Bonferroni/Holm's procedures). Therefore, the interpretation of results was performed with caution, based on the clinical significance.[48] For instance, current results echo previous findings, which similarly demonstrated infantile temperamental and parental competence issues in the CHD population.

Further, parenting stress trajectories were linearly presented, with "time" as a continuous measure. Non-linear analysis providing detailed information regarding the stress flexuosity, may highlight sensitive periods over infancy. The prenatal diagnosis, days and weeks around the surgery, and the early weeks at home have been described as over-stressful in the CHD population.[49] Parenting stress has been first measured at 3 months in our study, preventing us with any information regarding the stress around hospital discharge. Furthermore, many within the uni-ventricular heart infants undergo their second stage surgery around 4 to 6 months of age, which may be an additional stressful/traumatic event for the family after they have started adjusting to the situation at home. Families may benefit from earlier and longer assessment starting at hospital and throughout and beyond infancy, involving qualitative evaluations of participants' experiences.

8 | CONCLUSIONS

The current study novelty tracked parenting stress over infancy in a population of critically ill children and their parents. Findings indicate that the stress of parents of SV infants evoking around infant's temperamental characteristics, parental role restrictions, and attachment issues decreases with time. While parents of BV infants similarly experience attachment-related stress decrease, they also experience stress increase in relation to infant's distractibility. Parental support and interventions should be tailored to the trajectory of parental distress, the illness condition course, and typical characteristics. Interventions should focus on parental empowerment toward balanced coping and promotion of healthy parent-infant relationships.

CONFLICT OF INTEREST

The authors declare no conflict of interest.

AUTHOR CONTRIBUTIONS

Nadya Golfinshtein is the first and primary article author, and is responsible for study conceptualization and design, data analysis, statistics, and interpretation of results, article drafting and approval.

Alexandra Hanlon is the supervising biostatistician, and is responsible for data analysis and interpretation, critical revisions, and approval of article.

Janet Deatrick contributed to study conceptualization, interpretation of results, critical revisions, and approval of article.

Barbara Medoff-Cooper, the senior scientist, contributed to study conceptualization, interpretation of results, critical revisions, and approval of article.

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