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Posttraumatic stress and health-related quality of life in parents of children with cardiac rhythm devices

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Abstract

Purpose Studies have shown a high prevalence of post-traumatic stress disorders (PTSD) among parents of children with life-threatening diseases. However, it is yet unknown whether parents of children with cardiac rhythm device develop post-traumatic stress symptoms or even PTSD.

Methods This cross-sectional investigation is part of a comprehensive single-center study of long-term medical and psychosocial outcomes in pediatric patients with pacemaker (PM) and implantable cardioverter defibrillator (ICD). 69 patients (78%) were included in the study, with the participation of 69 mothers and 57 fathers. Parents completed the Posttraumatic Diagnostic Scale and Medical Outcomes Study Short Form-36 item questionnaire. Child's medical data was collected retrospectively from patients' hospital records.

Results At assessment, the patients (39% females) were on average 11.2 years old. The predominant device type was PM in 56 cases (81%). The mean time since device implantation was 6.3 years (SD = 4.3). Full heart-disease related PTSD was diagnosed in one mother and no father, while partial heart-disease-related PTSD was diagnosed in 3 mothers (4%) and 2 fathers (4%). Parental HRQoL—especially regarding the mental health dimension—was affected in both parents. In both parents, total PTSD symptom severity scores were a significant predictor for mental health summary scores after controlling for child age at implantation, presence of other non-cardiac disease in the child, parental age, and presence of own chronic disease.

Conclusions Special attention should be given to parental PTSD symptoms in the clinical follow-up of PM and ICD patients as some parents might probably benefit from psychological support.

Keywords Cardiology · Arrhythmia · Pacemaker · Implantable cardioverter defibrillator · Parents · Childhood

Introduction

Studies have shown a high prevalence of post-traumatic stress disorders (PTSD) among parents of children with life-threatening diseases [1–3]. PTSD is characterized by psychological reactions assigned to three symptom clusters (intrusion, avoidance, and hyperarousal) following exposure to a traumatic event. The presence of PTSD symptoms has been shown in parents of children with chronic heart disease [4–7]. Another study showed that 25% of the mothers and 26% of the fathers met the criteria for surgery-related PTSD after their child's open heart surgery [8]. However, only one study focused on parents of children with cardiac rhythm devices [9]. This study indicated no increased parental distress measured by the Brief Symptom Inventory [10], while PTSD was not considered. Thus, it is yet unknown whether parents of children with cardiac rhythm device develop post-traumatic stress symptoms or even PTSD based on the diagnostic criteria of the Diagnostic and Statistical Manual of Mental Disorders, 4th Edition (DSM-IV) [11].

Parents of children living with a pacemaker (PM) or implantable cardioverter defibrillator (ICD) face a variety of stressors related to their child's arrhythmia and medical

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treatment. These devices protect their children from life-threatening consequences of arrhythmia such as post-surgical atrioventricular block and sinus bradycardia, ventricular tachycardia, or fibrillation [12–14]. However, a large percentage of these parents has previously experienced fear of losing a child, or seen their child undergoing open heart surgery or other invasive treatments before cardiac rhythm device implantation. Health-related quality of life (HRQoL), a multidimensional concept integrating subjective perception of an individual's physical, psychological, and social health [15], has become an important and widely used outcome measure. Previous studies with parents of children with various congenital heart diseases (CHD) have indicated impaired parental HRQoL [8, 16–18]. To the best of our knowledge, no study has previously described HRQoL among parents of children with cardiac rhythm devices. Given that PTSD and affected HRQoL [15] might keep parents from supportive interactions with their child, it is important to know whether and how the parents are affected. Identifying affected parents allows to provide specific care, which may support the child's adjustment process to the disease.

The present study aims (1) to assess the prevalence of PTSD in the parents of children with PM or ICD. Furthermore, the study aims (2) to describe HRQoL in the patients' parents as compared to parents of sex- and age-matched healthy controls and to analyze maternal and paternal differences. Finally, we thought (3) to examine associations between parental HRQoL and child medical characteristics as well as associations between parental HRQoL and post-traumatic stress symptoms, while controlling for child medical characteristics (i.e., age at implantation, presence of other non-cardiac chronic disease) and parental sociodemographic characteristics (i.e., age, presence of own chronic disease).

Methods

Recruitment and inclusion criteria

We recruited parents whose children were treated between September 2015 and September 2016 at the cardiology outpatient clinic of the University Children's Hospital Zurich. Inclusion criteria were (1) child diagnosis of cardiac arrhythmia, (2) PM or ICD implantation > 3 months prior to study inclusion, (3) absence of Down syndrome or severe mental retardation in the child, (4) child age between 3 and 18 years during the recruiting period, and (5) fluency in German language of the caregiver. Exclusion criteria were permanent residency outside of Switzerland, refusal to sign the informed consent and device implantation within the last 3 months in order to minimize the influence of acute medical effects. Healthy controls were recruited via advertisements at the university and community day care centers (60%) and via best friends of the patients (40%) and were matched in child age and sex. Interviews and standardized questionnaires ensured that children of the control group had no chronic disease or cognitive impairments.

In total, 89 patients with PM and ICD were eligible for study participation. Twenty patients were excluded from the study for the following reasons: lack of time or interest ($n = 15$), lost to follow-up ($n = 4$), one family having two affected children ($n = 1$), which would differentiate them from all other families, especially with respect to the stress level. All remaining 69 patients (78%) were included in the study, with the participation of 69 mothers and 57 fathers.

This cross-sectional investigation is part of a comprehensive single-center study of long-term medical and psychosocial outcomes in pediatric patients with PM and ICD. The study was approved by the ethical review board of the Canton Zurich, Switzerland, and was performed in full accordance with the Declaration of Helsinki. All parents provided written informed consent after detailed explanation of study procedures and aims.

Measurements

Parents were asked to complete the questionnaires described below which were mailed to them. Child's medical data was collected retrospectively from patients' hospital records.

Child medical data

The time since initial device implantation (years) was calculated by the difference between the child's age at assessment and the age at initial device implantation. ICD patients were grouped according to whether they had previously experienced an aborted cardiac arrest or life threatening arrhythmia (secondary prevention) or not (primary prevention). Patients with structural CHD were categorized according to the complexity of the surgical repair into univentricular (i.e., Fontan type) and biventricular physiology. Previous open heart surgery and reanimation was indicated by yes vs. no. The number of device-related post-initial-implant surgeries was categorized into none, one, or more than one. Length of individual hospital stays related to cardiac disease was summed into the total length (days) of cardiac hospitalization. Current cardiac medication was assessed by the intake of heart failure and/or antiarrhythmic medication, dichotomized into yes or no. In addition, we indicated the presence of other non-cardiac chronic disease (e.g., neurological diseases with recurrent seizures).

Parental post-traumatic stress symptoms

PTSD was assessed by the validated German version of the Posttraumatic Diagnostic Scale (PDS) [19–21] which diagnoses PTSD according to DSM-IV [11] and rates PTSD symptom severity. The PDS includes four parts: (1) the type of traumatic event and, if there were multiple events, the most stressful one; (2) the time, circumstances, and emotions in the context of the trauma; (3) the frequency of 17 symptoms belonging to the three symptom clusters (intrusion, avoidance and hyperarousal) during the last month before assessment with a 4-point Likert scale (0–3). The 17 items are summarized by a total symptom severity score (0–51; score ≤ 10, mild severity; score ≥ 36, severe severity); and (4) impairments in different aspects of life. Full PTSD was diagnosed if parents reported at least one intrusion symptom, three avoidance symptoms, two hyperarousal symptoms with a duration of at least one month and impaired function in at least one life aspect [11]. For diagnosis of partial PTSD, parents had to report at least one symptom in each category, while the criteria with respect to the duration of the symptoms and impaired function were the same [21]. Internal consistency of the PTSD symptom severity score was good (for the mothers: Cronbach's $\alpha = .94$; for the fathers Cronbach's $\alpha = .91$).

Parental health-related quality of life

Self-reported HRQoL was assessed with the authorized German version of the Medical Outcomes Study Short Form-36 item questionnaire (SF-36) covering the 4 weeks before assessment [22, 23]. The SF-36 includes the following eight subscales: 'physical functions' (e.g., problems going upstairs), 'role physical' (e.g., problems at work due to physical problems), 'bodily pain' (e.g., how pain would restrict everyday activities), 'general health' (e.g., how general health would be rated compared to the last year), 'vitality' (e.g., low energy), 'social functions' (e.g., whether physical or emotional problems would influence the contact with friends), 'role emotional' (e.g., whether there are problems at work due to mental health problems) and 'mental health' (e.g., being unhappy). Each subscale ranges from 0 to 100 with higher scores indicating better HRQoL. The eight subscales can be summarized by two T-standardized component summary scores with a mean of 50 (SD = 10), i.e., a physical component summary (PCS) and a mental component summary (MCS) score. Internal consistency of all subscales was satisfactory to excellent (Table 3).

Life events within the family

The occurrence of major life events in the family during the 12 months prior to assessment was assessed by parental report using a list of the following 12 events [24]: birth of a child, divorce, marriage, person moving into the household, significant change in family income, indebtedness, removal, job change of either parent, unemployment of either parent, serious illness or accident of a family member, death of family member or a close friend, and a child's change of school. A life event score was computed by summing the number of life events (range 0–12).

Socio-economic status

Socio-economic status (SES) was assessed with a demographic questionnaire and estimated based on maternal education and paternal occupation ranging from 2 to 12, with 2 being the lowest and 12 the highest SES score. Three SES classes were assigned: lower (2–5), middle (6–9) and upper class (10–12). This measure has proven to be a valid indicator of SES in previous studies of the Swiss population [25]. Parental educational level was measured using a scale ranging from 1 to 6 points, with higher scores indicating higher level. Single parent was indicated by yes versus no.

Statistical analyses

Data were analyzed using SPSS 22.0 for Windows (SPSS, Inc., Chicago, IL, USA). All statistical tests were two-sided with a predefined significance level of $p < .05$. Cronbach's alpha was calculated for testing internal consistency of scale scores. Wilcoxon tests were performed to examine whether HRQoL scores of mothers and fathers differed significantly from the control group and to compare patient mothers' and fathers' scores from each other. For all comparisons, effect sizes by Cohen's d were computed (.20, small effect; .50, medium effect; > .80, large effect) [26]. Bivariate associations of parental HRQoL scores and studied variables were examined by means of Spearman and Kendall correlations due to non-normal distribution of many of the variables. Linear multiple regression models were used to predict HRQoL with, for both parents, the MCS as dependent variable. Normal plots of the residuals indicated certain deviations from normality. Because several standard transformations for the dependent variable did not substantially improve the distribution of the residuals we consider, from a distributional point of view, and also because of the moderate sample size, the linear regression as adequate. Because parental PCS was not decreased compared to the controls, no regression analysis was performed for this outcome variable. For both parents, five predictors were entered into the regression analyses: child age (years) at initial implantation, presence of other non-cardiac chronic disease in the child, parental age (years), presence of own chronic disease, and total PTSD symptom severity (score range 0–51). Selection of predictors was based on a priori hypotheses and bivariate correlations

with the dependent variable. The assumption of multicollinearity was tested by checking the correlation matrix (coefficients > .80) and the variance inflation factor (VIF).

Results

Child socio-demographic and medical characteristics

Table 1 shows the socio-demographic and medical characteristics of the children. At assessment, the patients (39% females) were on average 11.2 years old and belonged mostly to the middle or upper SES class (96%). The predominant device type was PM in 56 cases (81%), while 13 cases (19%) had an ICD. The mean time since device implantation was 6.3 years (SD = 4.3, range .4–17.9), with 6 children (9%) who received their device within the last 12 months. The majority of patients (65%) received their device before 6 years of age. Thirty of the 56 PM patients (54%) and 8 of the 13 ICD patients (62%) had a structural CHD and 31 of these 38 CHD patients (82%) had undergone open heart surgery. Twelve of the 69 patients (17%) suffered from an additional non-cardiac chronic disease: attention deficit hyperactivity disorder ($n = 2$), hearing disorder ($n = 1$), congenital hand or arm malformation ($n = 2$), diabetes mellitus ($n = 1$), neurological disease ($n = 5$), and diseases of the endocrine system ($n = 1$).

Table 1 Patient socio-demographic and medical characteristics ($n = 69$)

Age (years) at assessment	
M (SD), range	11.2 (4.6), 3.2–18.0
Female gender	27 (39%)
Socio-economic status	
High	17 (25%)
Middle	49 (71%)
Low	2 (3%)
Unknown	1 (1%)
Device type	
Pacemaker	56 (81%)
Implantable cardioverter defibrillator	13 (19%)
Age (years) at initial device implant	
Mean (SD), range	4.9 (4.4), 0–15.3
Newborn (age ≤ 28 days)	6 (9%)
Infant/toddler (29 days to 3 years)	21 (30%)
Preschool (3–6 years)	18 (26%)
School-age (6–13 years)	21 (30%)
Adolescent (age > 13 years)	3 (4%)
Electrophysiological disease	
Congenital heart block	24 (35%)
Postoperative heart block	19 (28%)
Sinoatrial node disease	13 (19%)
Ventricular tachycardia or fibrillation	13 (18%)
Implantable cardioverter defibrillator indication	
Primary prevention	7 (54%)
Secondary prevention	6 (46%)
Structural congenital heart disease	38 (55%)
Previous reanimation	9 (13%)
Number of device-related post-initial-implant surgeries	
Mean (SD), range	.7 (.8), 0–3
None	38 (55%)
One	19 (28%)
More than one	12 (17%)
Total length (days) of cardiac hospitalization, mean (SD), range	51.4 (58.0), 2–266
Current cardiac medication	20 (29%)
Other non-cardiac chronic disease	12 (17%)

Parental socio-demographic characteristics

The mean age of the mothers was 41.7 years (SD = 5.9, median = 42.0, range 28–60) and of the fathers 44.3 years (SD = 7.54, median = 44.0, range 28–68). In 66 cases (96%), the parents were biological parents while in three cases, the child was in care of adoptive parents which was in one case the grandmother only. Six mothers (9%) and 5 fathers (7%) were single parents. Eight mothers (12%) and 12 fathers (17%) had a university diploma. Five mothers (7%) and four fathers (6%) indicated to be affected by an own chronic physical disease (mothers: systemic lupus erythematosus, heart disease, asthma, diabetes, cancer; fathers: colitis ulcerosa, hereditary multiple exostoses, heart disease, diabetes), with one mother and one father also affected by a chronic heart disease (the mother also had an ICD). In addition, three fathers (5%) reported to be affected by an affective disorder. The majority of parents (80%) reported the occurrence of at least one major life event during the 12 months preceding the assessment. The death of a family member or a close friend (38%), serious illness/accident of a family member (28%), and a job change of either parent (33%) were the most frequently reported life events.

Parental post-traumatic stress disorders

Prevalence of post-traumatic stress symptoms and PTSD in the parents are shown in Table 2. Six mothers (13%) and 4 fathers (10%) met the criteria for full PTSD (due to any traumatic event). The child's heart disease was stated as a single and/or the most troubling traumatic event by 8 mothers (12%) and 4 fathers (6%). Of these, one mother (1%) met the criteria for full PTSD and 3 mothers (4%) for partial PTSD. None of the fathers met the criteria for full PTSD diagnosis due to their child's heart disease, while two fathers (4%) met the criteria for partial PTSD.

Table 2 Prevalence of post-traumatic stress symptoms and post-traumatic stress disorder (PTSD) according to DSM-IV in parents of children with cardiac rhythm devices

PDS items	Mothers ($n = 69$)	Fathers ($n = 57$)
Trauma exposure (any traumatic event)		
No trauma	20 (29%)	16 (28%)
One prior trauma	19 (28%)	22 (39%)
More than one prior trauma	29 (42%)	19 (33%)
Missings	1 (1%)	0
All 3 symptom clusters		
DSM-IV criterion for full PTSD met	6 (13%)	4 (10%)
DSM-IV criterion for partial PTSD met	10 (21%)	8 (21%)
Total symptom severity	4.5 (7.5), 0–33	3.5 (5.6), 0–25
Parents stating their child's heart disease as a traumatic event	8 (12%)	4 (6%)
Intrusion		
DSM-IV criterion met	5 (7%)	4 (7%)
Number of symptoms	2.9 (2.5), 0–6	3.0 (1.4), 1–4
Avoidance		
DSM-IV criterion met	1 (1%)	0
Number of symptoms	2.3 (3.2), 0–8	1.0 (.8), 0–2
Hyperarousal		
DSM-IV criterion met	4 (5%)	1 (2%)
Number of symptoms	2.1 (1.8), 0–5	.3 (.5), 0–1
All 3 symptom clusters		
DSM-IV criterion for full PTSD met	1 (1%)	0
DSM-IV criterion for partial PTSD met	3 (4%)	2 (4%)
Total symptom severity	4.5 (7.5), 0–33	5.3 (3.0), 2–9

PTSD post-traumatic stress disorders, *DSM-IV* Diagnostic and Statistical Manual of Mental Disorders, 4th Edition

Parental health-related quality of life

HRQoL mean values of the mothers and fathers and statistics for comparison with mothers, respectively, fathers of the control group as well as between mothers and fathers of the patient sample are presented in Table 3. With exception of the subscale 'role emotional', all maternal-reported HRQoL subscale scores were significantly lower than the control group. For the patient's fathers, only the subscale 'mental health' showed a decrease compared to the control group. For both parents, the largest effect between the patient and control group was found for 'mental health'. The MCS also differed between the patient and control group with medium effect size in both parents, while no significant differences were found for PCS. Patient parents had lower mental health than the control parents. In the patient group, no significant differences were found between maternal- and paternal-reported subscales or summary scores.

Associations between parental HRQoL and child socio-demographic and medical characteristics

Parental MCS scores were not significantly associated with child age at assessment, gender, and SES (Table 4). With exception of two variables, maternal- and paternal-reported MCS scores were not significantly associated with child medical characteristics, i.e., time since initial device implantation, device type, structural CHD, number of device-related post-initial-implant surgeries, previous reanimation, total length of cardiac hospitalization, and need for cardiac medication. Among the mothers, lower mental health was

Table 3 HRQoL of mothers and fathers with children with pacemaker and implantable cardioverter defibrillator: comparison with control group and between mothers and fathers

SF-36 scales	Mothers of the patient group		Mothers of the control group		Fathers of the patient group		Fathers of the control group		Comparison: patient–control group (mothers)		Comparison: patient–control group (fathers)		Comparison: mothers–fathers (patient group)	
	n	*M* (SD)	*n*	*M* (SD)	*n*	*M* (SD)	*n*	*M* (SD)	p^a	d^b	p^a	d^b	p^a	d^b
Physical functions (.92[c]/.95[d])	69	93.6 (14.0)	67	98.2 (4.0)	57	92.1 (18.3)	51	96.7 (5.5)	**.006**	− .45	.90	− .34	.39	.09
Role physical (.88[c]/.93[d])	69	92.8 (22.3)	67	98.9 (5.2)	57	88.6 (29.2)	51	94.6 (18.2)	**.031**	− .38	.35	− .25	.30	.16
Bodily pain (.77[c]/.86[d])	69	83.8 (21.0)	67	90.2 (14.8)	57	83.3 (22.6)	51	89.6 (16.3)	**.041**	− .35	.44	− .32	.60	.02
General health (.76[c]/.75[d])	69	78.9 (15.5)	67	85.7 (12.4)	57	77.5 (17.7)	51	78.9 (16.4)	**.012**	− .48	.65	− .08	.76	.08
Vitality (.83[c]/.78[d])	69	61.5 (16.3)	67	67.5 (14.4)	57	61.5 (15.3)	51	67.1 (16.1)	**.026**	− .39	.09	− .36	.63	.00
Social functions (.82[c]/.73[d])	69	85.9 (18.3)	67	93.5 (13.3)	57	90.4 (14.9)	51	94.4 (16.4)	**.010**	− .48	.18	− .26	.15	− .27
Role emotional (.65[c]/.85[d])	68	90.2 (22.3)	67	92.0 (21.0)	57	90.6 (25.8)	51	94.8 (20.4)	.732	− .08	.72	− .18	.75	− .02
Mental health (.75[c]/.76[d])	69	71.4 (11.2)	67	80.4 (11.9)	57	70.4 (11.7)	51	81.4 (12.5)	**<.001**	− .78	**<.001**	− .91	.80	.09
PCS	68	54.7 (7.2)	67	56.8 (3.7)	57	53.9 (8.4)	51	54.8 (3.9)	.078	− .37	.67	− .14	.50	.10
MCS	68	48.7 (7.5)	67	52.1 (6.8)	57	49.5 (6.9)	51	53.2 (7.4)	**.009**	− .47	**.005**	− .52	.19	− .11

PCS physical component summary score, *MCS* mental component summary score

Higher scores indicate better HRQoL

[a]Wilcoxon-Test was performed

[b]*d* = effect size according to Cohen (.20, small effect; .50 medium effect; > .80, large effect) [1]

[c]Cronbach's alpha for the mothers

[d]Cronbach's alpha for the fathers

associated with a younger child age at implantation. Among the fathers, MCS scores improved with the presence of other non-cardiac chronic diseases in the child.

Associations between parental HRQoL and parental socio-demographic characteristics

In both parents, better mental health measured by the MCS was significantly associated with lower PTSD symptom severity. In addition, an older age of the mothers was significantly associated with better maternal-reported MCS scores; paternal-reported MCS scores were significantly correlated with the presence of an own chronic disease. Table 5 summarizes the statistics for two separate linear multiple regression models with parental MCS scores as dependent variable. Each overall multivariate model was significant and accounted for 23% in the mothers, respectively, 49% in the fathers of the total variance. In both parents, the MCS was significantly associated with posttraumatic stress symptoms, while maternal-reported MCS scores were also predicted by child age at initial implantation and paternal-reported MCS scores were also predicted by the presence of an own chronic disease.

Discussion

This cross-sectional study provides a comprehensive insight into self-reported PTSD and HRQoL in parents of children with PM or ICD. Our results show that full heart-disease-related PTSD based on the Diagnostic and Statistical Manual of Mental disorder, 4th edition (DSM-IV), was diagnosed in one mother and no father, while partial heart-disease-related PTSD was diagnosed in 3 mothers and 2 fathers. Parental HRQoL—especially regarding the mental health dimension of HRQoL—is affected in both parents.

The prevalence of heart-disease related PTSD in the parents of this sample is considerable lower compared to other studies describing PTSD or acute stress disorders in parents of children with various heart diseases [4–7] (83%, Cantwell-Bartl 2013; 18–34%, Franich-Ray 2013). Since parental post-traumatic stress depends on the time since cardiac surgery [27, 28], it is possible that the period of 6.3 years since child's device implantation offered sufficient time to accommodate. The families were also mainly from middle or upper SES class which might have influenced the PTSD prevalence. A higher SES normally implies more financial and social resources, which can serve as protective factors in stressful situations [29]. But still, a small proportion of parents experienced post-traumatic stress symptoms due to their child's heart disease in the long-term. However, no significant group difference

Table 4 Correlations of parental HRQoL scores with child socio-demographic characteristics, child medical characteristics and parental characteristics

	Mothers (n = 64–68) MCS	Fathers (n = 54–57) MCS
Child socio-demographic characteristics		
Age (years) at assessment[a]	.17	.07
Female gender[b]	− .06	− .27
SES[a]	− .02	− .16
Child medical characteristics		
Age (years) at initial device implantation[a]	.27*	.11
Time (years) since initial device implantation[a]	− .07	− .03
Device type[b]	− .12	− .07
Structural congenital heart disease[b]	− .09	− .05
Number of device-related post-initial-implant surgeries[a]	− .14	− .07
Previous reanimation[b]	− .09	.08
Total length (days) of cardiac hospitalization[a]	− .15	− .06
Current cardiac medication[b]	.05	− .02
Other non-cardiac chronic disease[b]	.17	.28*
Parental characteristics		
Age (years)[a]	.18*	.02
Educational level[a]	− .04	− .12
Single parent[b]	.09	.02
Presence of own chronic disease[c]	.22	− .39**
Number of previous life events[a]	− .08	− .08
Total PTSD symptom severity[a]	− .29**	− .30**

SES socio-economic status, *MCS* mental component summary score

*$p \leq .05$, **$p \leq .01$, ***$p \leq .001$

[a]Spearman correlation coefficients are presented; [b] Kendall correlation coefficients are presented

[c]Defined by the presence of a physical or psychological disease

Table 5 Summary of multiple regression analysis with parental MCS scores as dependent variables

Predictors	Mothers (n = 64)				Fathers (n = 54)			
	MCS				MCS			
	B	SE B	β	p	B	SE B	β	p
Child age (years) at initial implantation	.41	.19	.24	**.04**	.12	.15	.09	.43
Other non-cardiac chronic disease	3.59	2.38	.18	.14	3.52	1.81	.21	.06
Parental age (years)	.03	.15	.02	.85	.02	.09	.02	.87
Presence of own chronic disease[a]	3.56	3.27	.13	.28	− 6.76	2.59	− .29	**.01**
Total PTSD symptom severity	− .49	.13	− .43	**<.001**	− .62	.13	− .50	**<.001**
	F (5) = 5.47, p < .001 R^2 = .23, $R^2_{adj.}$ = .26				F (5) = 9.32, p < .001 R^2 = .49, $R^2_{adj.}$ = .44			

MCS mental component summary score, *PTSD* post-traumatic stress disorders, *B* regression coefficient, *SE B* standard error of the regression coefficient, β standardized regression coefficient

[a]Defined by the presence of a physical or psychological disease

between parents of children with PM vs. ICD regarding the PTSD prevalence was found in our study (data not shown). The PTSD prevalence due to any traumatic event was 13% in the mothers and 10% in the fathers which is higher than in the general European population ranging from .6 to 6.7% [30]. Thus, it might be that parents of children with PM or ICD might have the tendency to cope in a more traumatic way with stressful events than parents of healthy children.

Based on a HRQoL study with parents of children after open heart surgery [8], on studies with parents of children with various heart diseases [16–18] and studies showing

decreased HRQoL in the patients themselves [9, 31–37], lower HRQoL was expected in the parents of PM or ICD patients. In line with our hypothesis, parental HRQoL—especially regarding the mental health dimension of HRQoL—is affected in both parents. This is in contrast to the single study including parents of children with cardiac rhythm devices evaluating their distress measured by the Brief Symptom Inventory [9]. However, the children in our study had a younger age at implantation and thus including more challenges [9] which might have contributed to this difference. Furthermore, we analyzed the extent to which child socio-demographic and medical characteristics are associated with parental HRQoL. While child socio-demographic characteristic did not correlate, better mental health as measured by the MCS was associated with an older child age at implantation among the mothers and with the presence of other non-cardiac chronic diseases in the child among the fathers. After multivariate analysis, only the child's age at initial implantation (among the child medical characteristics) turned out to be a significant predictor for maternal MCS scores. Thus, we were not able to show a significant association between parental HRQoL and the presence of a structural CHD or previous open heart surgery in the child in our study. Also no significant HRQoL difference between the mothers and fathers of children with PM or ICD was found. This might be explained by the same reasons as mentioned above.

In our study, we also attempted to identify other factors that are related to parental HRQoL. In both parents, PTSD symptom severity scores (due to any traumatic event) were significant predictors for parental HRQoL (mental health dimension) after controlling for other study variables such as child age at initial implantation, presence of other non-cardiac chronic disease in the child, parental age, and presence of own chronic disease. Better mental health was associated with fewer PTSD symptoms, while in the fathers the presence of own chronic disease was also significant predictor. This might be related to the fact that 3 fathers reported to be affected by an affective disorder. Thus, special attention should be given to parental PTSD symptoms in the clinical follow-up of PM and ICD patients as some parents might probably benefit from psychological support.

The strengths of our study include the use of standardized multidimensional questionnaires and the comparison of our HRQoL results with control data. Furthermore, a high proportion of fathers (83% of participating families) participated in our study, allowing comparisons between maternal and paternal PTSD and HRQoL data. Our data originates from the cardiology outpatient clinic of the University Children's Hospital Zurich which is responsible for about 45% of the whole Swiss population. The device therapy of cardiac arrhythmia patients relies on published guidelines [12–14]. Thus, the results of our study cannot be transferred to patients treated in other centers that are not in line with these guidelines, but can be transmitted with caution to all patients in Switzerland and to patients living in another Central European country with similar social and economic structure. In terms of a post hoc power analysis ($\alpha = .05$, two-tailed) using the G*power software [38], our sample size provided adequate power to detect moderate effect sizes within the multiple regression analysis. Nevertheless, our study has several limitations. First, only a rather small number of pediatric patients with cardiac rhythm devices, especially patients with ICD, was included, making it difficult to compare parental post-traumatic stress symptoms or HRQoL with respect to the device type. Secondly, the majority of families belonged to the middle or upper SES class. Thus, our results are not representative for the whole population. Most probably, the following two reasons might have caused this bias: a) Non-German speaking parents were excluded from the study participation, and b) among eligible families, those from lower SES classes might have declined participation because of fewer resources to cope with the child's disease. Thus, we cannot rule out that our findings may be biased in an overly positive direction. Also we cannot rule out the possibility that parents who participated in our study differed with respect to parents' age, educational level or other variables like for example HRQoL from those who did not. However, a dropout-analysis with child medical data (data not shown) revealed no significant differences between the participants and non-participants regarding child sex, child age at study beginning, child age at initial device implantation, device type, the presence of structural congenital heart disease and Swiss nationality. Third, the cross-sectional design of our study does not allow for any causal relationships. Therefore, we do not know whether low HRQoL contributes to the development of PTSD or PTSD leads to low HRQoL in the parents. Fourth, the fact that our multivariate model explained 23% in the mothers, respectively, 49% in the fathers of the variance in MCS scores indicates that the MCS of both parents is not predicted by the same variables. Future studies might address this gender difference in more detail. Also other variables, not assessed in this study as for example social support or parents' personality traits, which might also influence parental HRQoL, might be included in a future study. Fifth, the SF-36 measures HRQoL by two standardized component summary scores: physical health and mental health. In our study, talking about mental health, we always meant the HRQoL component 'meant health' (MCS) assessed by the SF-36 and not mental health in the whole spectrum as a clinical interview would capture. Sixth, we were not able to provide PTSD prevalence rates according to DSM-V because no updated and validated German version of the PDS was available at the time of data collection.

An important issue for future research is to perform multi-center and prospective cohort studies with parents of children with PM or ICD. A recent systematic review has shown that this population is marginally studied [39]. With regard to clinical practice, our results suggest that addressing post-traumatic stress symptoms and HRQoL in the parents might be beneficial. Even though a relatively small number of parents showed child heart-disease-related PTSD in the long term, the PTSD prevalence due to any traumatic event was as high as 13% in mothers and 10% in the fathers. This significantly influences parental HRQoL and might keep parents from supportive interactions with their child which, in turn, place the child at increased risk of psychological adjustment problems [40, 41]. A recent review about psychological interventions designed to change parent cognition, behavior, or both for parents of children with chronic illnesses have shown that psychological interventions led to improved adaptive parenting behavior and improved parental mental health [42]. Thus, providing more integrated care at the hospital might be very helpful for the affected families [16, 43, 44].

In conclusion, the results of our study showed that parental HRQoL is reduced compared to controls. Furthermore, a few parents suffered from full or partial PTSD even though that the PTSD prevalence due to their child's heart disease was small. Thus, special attention should be given to parental PTSD symptoms in the clinical follow-up of PM and ICD patients as some parents might benefit from psychological support.

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References

1. Stuber, M. L., & Shemesh, E. (2006). Post-traumatic stress response to life-threatening illnesses in children and their parents. *Child and Adolescent Psychiatric Clinics of North America, 15*(3), 597–609. https://doi.org/10.1016/j.chc.2006.02.006.
2. Melnyk, B. M., Alpert-Gillis, L., Feinstein, N. F., Crean, H. F., Johnson, J., Fairbanks, E., et al. (2004). Creating opportunities for parent empowerment: Program effects on the mental health/coping outcomes of critically ill young children and their mothers. *Pediatrics, 113*(6), E597–E607. https://doi.org/10.1542/peds.113.6.e597.
3. Nelson, L. P., & Gold, J. I. (2012). Posttraumatic stress disorder in children and their parents following admission to the pediatric intensive care unit: A review. *Pediatric Critical Care Medicine, 13*(3), 338–347. https://doi.org/10.1097/PCC.0b013e3182196a8f.
4. Ernst, M. M., Marino, B. S., Cassedy, A., Piazza-Waggoner, C., Franklin, R. C., Brown, K., et al. (2018). Biopsychosocial predictors of quality of life outcomes in pediatric congenital heart disease. *Pediatric Cardiology, 39*(1), 79–88. https://doi.org/10.1007/s00246-017-1730-6.
5. Franich-Ray, C., Bright, M. A., Anderson, V., Northam, E., Cochrane, A., Menahem, S., et al. (2013). Trauma reactions in mothers and fathers after their infant's cardiac surgery. *Journal of Pediatric Psychology, 38*(5), 494–505. https://doi.org/10.1093/jpepsy/jst015.
6. Cantwell-Bartl, A. M., & Tibballs, J. (2013). Psychosocial experiences of parents of infants with hypoplastic left heart syndrome in the PICU. *Pediatric Critical Care Medicine, 14*(9), 869–875. https://doi.org/10.1097/PCC.0b013e31829b1a88.
7. Helfricht, S., Latal, B., Fischer, J. E., Tomaske, M., & Landolt, M. A. (2008). Surgery-related posttraumatic stress disorder in parents of children undergoing cardiopulmonary bypass surgery: A prospective cohort study. *Pediatric Critical Care Medicine, 9*(2), 217–223. https://doi.org/10.1097/PCC.0b013e318166eec3.
8. Landolt, M. A., Buechel, E. V., & Latal, B. (2011). Predictors of parental quality of life after child open heart surgery: A 6-month prospective study. *Journal of Pediatrics, 158*(1), 37–43. https://doi.org/10.1016/j.jpeds.2010.06.037.
9. Webster, G., Panek, K. A., Labella, M., Taylor, G. A., Gauvreau, K., Cecchin, F., et al. (2014). Psychiatric functioning and quality of life in young patients with cardiac rhythm devices. *Pediatrics, 133*(4), e964–e972. https://doi.org/10.1542/peds.2013-1394.
10. Derogatis, L. R., & Melisaratos, N. (1983). The Brief Symptom Inventory: An introductory report. *Psychological Medicine, 13*(3), 595–605.
11. American-Psychiatric-Association. (1994). *The diagnostic and statistical manual of mental disorders*. Washington, DC: American Psychiatric Association.
12. Koenig, P., Hijazi, Z. M., & Zimmerman, F. (2004). *Essential pediatric cardiology*. New York: McGraw-Hill Medical Publishing Division.
13. Epstein, A. E., DiMarco, J. P., Ellenbogen, K. A., Estes, N. A., 3rd, Freedman, R. A., Gettes, L. S., et al. (2008). ACC/AHA/HRS 2008 Guidelines for Device-Based Therapy of Cardiac Rhythm Abnormalities: A report of the American College of Cardiology/American Heart Association Task Force on Practice Guidelines (Writing Committee to Revise the ACC/AHA/NASPE 2002 Guideline Update for Implantation of Cardiac Pacemakers and Antiarrhythmia Devices) developed in collaboration with the American Association for Thoracic Surgery and Society of Thoracic Surgeons. *Journal of the American College of Cardiology, 51*(21), e1–e62. https://doi.org/10.1016/j.jacc.2008.02.032.
14. Epstein, A. E., DiMarco, J. P., Ellenbogen, K. A., Estes, N. A., 3rd, Freedman, R. A., Gettes, L. S., et al. (2013). 2012 ACCF/AHA/HRS focused update incorporated into the ACCF/AHA/HRS 2008 guidelines for device-based therapy of cardiac rhythm abnormalities: A report of the American College of Cardiology Foundation/American Heart Association Task Force on Practice Guidelines and the Heart Rhythm Society. *Journal of the American College of Cardiology, 61*(3), e6–e75.
15. World-Health-Organization. (1948). *Constitution of the World Health Organization basic document*. Geneva: World-Health-Organization.
16. Eagleson, K. J., Justo, R. N., Ware, R. S., Johnson, S. G., & Boyle, F. M. (2013). Health-related quality of life and congenital heart disease in Australia. *Journal of Paediatrics and Child Health, 49*(10), 856–864. https://doi.org/10.1111/jpc.12296.
17. Lawoko, S., & Soares, J. J. (2003). Quality of life among parents of children with congenital heart disease, parents of children with other diseases and parents of healthy children. *Quality of Life Research, 12*(6), 655–666.
18. Arafa, M. A., Zaher, S. R., El-Dowaty, A. A., & Moneeb, D. E. (2008). Quality of life among parents of children with heart

disease. *Health and Quality of Life Outcomes, 6,* 91. https://doi.org/10.1186/1477-7525-6-91.
19. Foa, E. B., Cashman, L., Jaycox, L., & Perry, K. (1997). The validation of a self-report measure of posttraumatic stress disorder: The Posttraumatic Diagnostic Scale. *Psychological Assessment, 9*(4), 445–451. https://doi.org/10.1037//1040-3590.9.4.445.
20. Griesel, D., Wessa, M., & Flor, H. (2006). Psychometric qualities of the German version of the Posttraumatic Diagnostic Scale (PTDS). *Psychological Assessment, 18*(3), 262–268. https://doi.org/10.1037/1040-3590.18.3.262.
21. Stein, M. B., Walker, J. R., Hazen, A. L., & Forde, D. R. (1997). Full and partial posttraumatic stress disorder: Findings from a community survey. *American Journal of Psychiatry, 154*(8), 1114–1119.
22. Ware, J., Snow, K., & Kosinski, M. (2000). *SF-36 Health Survey: Manual and interpretation guide*. Lincoln, RI: Quality Metric Inc.
23. Bullinger, M., & Kirchberger, I. (1998). *SF-36 Fragebogen zum Gesundheitszustand. Handanweisung*. Göttingen: Hogrefe.
24. Landolt, M. A., & Vollrath, M. (1998). *Life event scale*. Zurich: University Childern's Hospital Zurich.
25. Largo, R. H., Pfister, D., Molinari, L., Kundu, S., Lipp, A., & Duc, G. (1989). Significance of prenatal, perinatal and postnatal factors in the development of AGA preterm infants at five to seven years. *Developmental Medicine and Child Neurology, 31*(4), 440–456.
26. Cohen, J. (1992). A power primer. *Psychological Bulletin, 112*(1), 155–159. https://doi.org/10.1037/0033-2909.112.1.155.
27. Kazak, A. E., Kassam-Adams, N., Schneider, S., Zelikovsky, N., Alderfer, M. A., & Rourke, M. (2006). An integrative model of pediatric medical traumatic stress. *Journal of Pediatric Psychology, 31*(4), 343–355. https://doi.org/10.1093/jpepsy/jsj054.
28. Price, J., Kassam-Adams, N., Alderfer, M. A., Christofferson, J., & Kazak, A. E. (2016). Systematic review: A reevaluation and update of the integrative (trajectory) model of pediatric medical traumatic stress. *Journal of Pediatric Psychology, 41*(1), 86–97. https://doi.org/10.1093/jpepsy/jsv074.
29. Trickey, D., Siddaway, A. P., Meiser-Stedman, R., Serpell, L., & Field, A. P. (2012). A meta-analysis of risk factors for post-traumatic stress disorder in children and adolescents. *Clinical Psychology Review, 32*(2), 122–138. https://doi.org/10.1016/j.cpr.2011.12.001.
30. Wittchen, H. U., Jacobi, F., Rehm, J., Gustavsson, A., Svensson, M., Jonsson, B., et al. (2011). The size and burden of mental disorders and other disorders of the brain in Europe 2010. *European Neuropsychopharmacology, 21*(9), 655–679. https://doi.org/10.1016/j.euroneuro.2011.07.018.
31. da Silva, K. R., Costa, R., De Oliveira, R. M., Lacerda, M. S., Huang, A. I. U., Rossi, M. B., et al. (2013). Quality of life and functional capacity after long-term right ventricular pacing in pediatrics and young adults with congenital atrioventricular block. *Pacing and Clinical Electrophysiology, 36*(12), 1539–1549. https://doi.org/10.1111/pace.12258.
32. Czosek, R. J., Bonney, W. J., Cassedy, A., Mah, D. Y., Tanel, R. E., Imundo, J. R., et al. (2012). Impact of cardiac devices on the quality of life in pediatric patients. *Circulation: Arrhythmia and Electrophysiology, 5*(6), 1064–1072. https://doi.org/10.1161/CIRCEP.112.973032.
33. Gaies, M. G., Watnick, C. S., Gurney, J. G., Bove, E. L., & Goldberg, C. S. (2011). Health-related quality of life in patients with congenitally corrected transposition of the great arteries. *Journal of Thoracic and Cardiovascular Surgery, 142*(1), 136–141. https://doi.org/10.1016/j.jtcvs.2010.11.035.
34. Cheng, P., Gutierrez-Colina, A. M., Loiselle, K. A., Strieper, M., Frias, P., Gooden, K., et al. (2014). Health related quality of life and social support in pediatric patients with pacemakers. *Journal of Clinical Psychology in Medical Settings, 21*(1), 92–102. https://doi.org/10.1007/s10880-013-9381-0.
35. Sears, S. F., Hazelton, A. G., St Amant, J., Matchett, M., Kovacs, A., Vazquez, L. D., et al. (2011). Quality of life in pediatric patients with implantable cardioverter defibrillators. *American Journal of Cardiology, 107*(7), 1023–1027. https://doi.org/10.1016/j.amjcard.2010.11.027.
36. Costa, R., da Silva, K. R., Mendonca, R. C., Nishioka, S., Siqueira, S., Tamaki, W. T., et al. (2007). Incidence of shock and quality of life in young patients with implantable cardioverter-defibrillator. *Arquivos Brasileiros de Cardiologia, 88*(3), 258–264. https://doi.org/10.1590/S0066-782x2007000300002.
37. DeMaso, D. R., Lauretti, A., Spieth, L., van der Feen, J. R., Jay, K. S., Gauvreau, K., et al. (2004). Psychosocial factors and quality of life in children and adolescents with implantable cardioverter-defibrillators. *American Journal of Cardiology, 93*(5), 582–587. https://doi.org/10.1016/j.amjcard.2003.11.022.
38. Faul, F., Erdfelder, E., Lang, A. G., & Buchner, A. (2007). G*Power 3: A flexible statistical power analysis program for the social, behavioral, and biomedical sciences. *Behavior Research Methods, 39*(2), 175–191. https://doi.org/10.3758/Bf03193146.
39. Pyngottu, A., Werner, H., Lehmann, P., & Balmer, C. (2019). Health-related quality of life and psychological adjustment of children and adolescents with pacemakers and implantable cardioverter defibrillators: A systematic review. *Pediatric Cardiology, 40*(1), 1–16. https://doi.org/10.1007/s00246-018-2038-x.
40. Hovels-Gurich, H. H., Konrad, K., Skorzenski, D., Minkenberg, R., Herpertz-Dahlmann, B., Messmer, B. J., et al. (2007). Long-term behavior and quality of life after corrective cardiac surgery in infancy for tetralogy of Fallot or ventricular septal defect. *Pediatric Cardiology, 28*(5), 346–354. https://doi.org/10.1007/s00246-006-0123-z.
41. Landolt, M. A., Ystrom, E., Stene-Larsen, K., Holmstrom, H., & Vollrath, M. E. (2014). Exploring causal pathways of child behavior and maternal mental health in families with a child with congenital heart disease: A longitudinal study. *Psychological Medicine, 44*(16), 3421–3433. https://doi.org/10.1017/S0033291713002894.
42. Eccleston, C., Fisher, E., Law, E., Bartlett, J., & Palermo, T. M. (2015). Psychological interventions for parents of children and adolescents with chronic illness. *Cochrane Database Systematic Reviews*. https://doi.org/10.1002/14651858.cd009660.pub3.
43. Brosig, C., Yang, K., Hoffmann, R. G., Dasgupta, M., & Mussatto, K. (2014). The role of psychology in a pediatric outpatient cardiology setting: Preliminary results from a new clinical program. *Journal of Clinical Psychology in Medical Settings, 21*(4), 337–346. https://doi.org/10.1007/s10880-014-9404-5.
44. Doherty, N., & Utens, E. (2016). A family affair. In C. McCusker & F. Casey (Eds.), *Congenital heart disease and neurodevelopment. Understanding and improving outcomes* (pp. 107–115). Amsterdam: Elsevier.