



Brief Report

# Perceived Parental Style Is Better in Adults with Congenital Heart Disease than Healthy Controls—But There Is Work Left to Do in Specific Subgroups

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**Abstract:** Objective: To compare perceived parental style in a large cohort of adults with congenital heart disease (ACHD) to healthy reference (RCs). Furthermore, factors associated with perceived parental style were determined in ACHD. Patients and Methods: From September 2016 to April 2019, 912 ACHD (34.9 ± 10.4 years, 45% female) and 175 RCs (35.8 ± 12.2 years, 53% female) completed the Measure of Parental Style (MOPS) questionnaire. Results: After adjusting for age and sex, ACHD recalled the parental style of both their parents to be significantly less indifferent (mother: ACHD: 1.2 ± 0.01 vs. RC: 1.3 ± 0.03,  $p < 0.001$ ; father: ACHD: 1.3 ± 0.02 vs. RC: 1.7 ± 0.05,  $p < 0.001$ ), overcontrolling (mother: ACHD: 1.6 ± 0.63 vs. RC: 1.9 ± 0.62,  $p < 0.001$ ; father: ACHD: 1.4 ± 0.52 vs. RC: 1.5 ± 0.50,  $p < 0.001$ ), and abusive (mother: ACHD: 1.2 ± 0.47 vs. RC: 1.4 ± 0.46,  $p < 0.001$ ; father: ACHD: 1.3 ± 0.59 vs. RC: 1.5 ± 0.57,  $p < 0.001$ ) than healthy controls did. In ACHD, female sex ( $\beta = 0.068$ ,  $p = 0.017$ ), higher age ( $\beta = 0.005$ ,  $p = 0.003$ ), Ebstein anomaly ( $\beta = 0.170$ ,  $p = 0.005$ ), and cyanotic CHD ( $\beta = 0.336$ ,  $p = 0.004$ ) contribute to perceiving the parental style of at least one of the parents negatively. Conclusions: While ACHD appear to recall the parental style to be less negative, subgroup analysis revealed specific patients at risk. These findings point to the need for interventions in specific subgroups susceptible to psychological distress.

**Keywords:** congenital heart disease; parental bonding; psychological; mental health



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## 1. Introduction

Medical aftercare for patients with congenital heart diseases (CHD) nowadays also needs to consider psychological aspects of mental health [1]. While quality of life and sense of coherence appear to generally be good [2,3], the prevalence of psychological illnesses, such as depression or mood and anxiety disorders, was found to be increased in adults with CHD (ACHD) [4,5]. These findings are especially alarming, considering depression is a major risk factor for adverse cardiovascular disease (CVD) outcomes and increased mortality [6].

In light of this connection between CVD and depression, together with the increased prevalence of psychological disorders in ACHD, undetected symptoms of depressive disorders might worsen the already enhanced risk for mortality in CHD patients [7,8]. And this does not even take into account the emotional stressors induced by growing up and living with CHD since childhood [8,9].

One risk factor for the development of psychological illnesses can be found in dysfunctional parenting styles. Patients with depression, anxiety disorder, and bulimia all

report having lived through dysfunctional parenting styles, even compared to healthy controls [10]. In addition to these clinical conditions, negatively perceived parenting styles have also been found to be associated with depression and other mental disorders in secondary school students [11] and hospitalization and suicide attempts in substance abusers [12] and in military-enlisted men [13]. While depression and anxiety have been studied considerably in CHD patients [8], the current literature on parental style is rather sparse, limited to adolescents, and inconsistent [14,15].

Therefore, this study aimed to compare, for the first time, perceived parental style, quantified by the Measure of Parental Style (MOPS) questionnaire, in a large cohort of ACHD to healthy controls. Furthermore, factors associated with a negatively perceived parental style were determined in ACHD.

## 2. Patients and Methods

### 2.1. Study Subjects

From September 2016 to January 2019, 912 patients with various CHD ( $34.9 \pm 10.4$  years, range: 20.1–69.4 years; 45% female) completed a German version of the “Measure of Parental Style” (MOPS) questionnaire [16] during their routine follow-up at the outpatient department of the German Heart Center Munich. All patients were free of any neurological diseases. CHD severity, according to ACC criteria [17], was distributed as follows: 482 complex, 317 moderate, and 113 simple.

For comparisons, 175 healthy controls ( $35.8 \pm 12.2$  years, range: 20.8–69.8 years, 53% female) were surveyed online in April 2019. Detailed information on the study subgroups is given in Table 1.

**Table 1.** Study subjects.

	n	Sex (Female) n (%)	Age (Years) Mean $\pm$ SD
<b>Congenital Heart Disease</b>	912	413 (45%)	$34.9 \pm 10.4$
<b>Reference Cohort</b>	175	92 (53%)	$35.8 \pm 12.2$
<i>p</i> -value *	-	0.092	0.348
<b>Aortic Stenosis</b>	142	36 (25%)	$33.7 \pm 9.6$
<b>Tetralogy of Fallot</b>	231	105 (45%)	$35.1 \pm 10.4$
<b>TGA after Arterial Switch</b>	47	16 (34%)	$24.9 \pm 3.7$
<b>TGA after Rastelli and CCTGA</b>	55	26 (47%)	$33.6 \pm 10.6$
<b>TGA Senning/Mustard</b>	95	29 (31%)	$36.4 \pm 5.8$
<b>Isolated Shunts</b>	102	67 (65%)	$36.1 \pm 11.2$
<b>Fontan Circulation</b>	49	20 (41%)	$32.1 \pm 6.4$
<b>Ebstein Anomaly</b>	93	64 (69%)	$40.5 \pm 13.0$
<b>Cyanotic Native or Palliated</b>	20	12 (60%)	$40.9 \pm 10.8$
<b>Coarctation of the Aorta</b>	78	38 (49%)	$34.6 \pm 11.1$

\* significant with  $p < 0.05$ .

All subjects gave written informed consent after being provided with information about the study protocol. The study was conducted in accordance with the Declaration of Helsinki (revised 2008) and approved by the local ethical board of the Technical University of Munich.

### 2.2. Measure of Parental Style

The “Measure of Parental Style” (MOPS) [16] is an internationally recognized self-assessment questionnaire of perceived parental style. The assessment consists of an identical mother and father version with 15 items. Each item is phrased as a “description of

their mother's and their father's behavior toward the participant in their first 16 years of life, and is answered on a 4-item Likert scale (1 = "does not apply at all" to 4 = "applies very much"). The mean of all ratings was calculated to attain a total score, according to Rumpold and colleagues [16]. Aside from the total score, the items can also be expressed in a three-factor solution showing the subscales "Indifference", "Over-control" and "Abuse". Again, following Rumpold et al. [16], "Indifference" was calculated as the mean of items 5, 8, 10, 11, 12, 13, "Over-control" as the mean of items 1, 3, 4, 6, and "Abuse" as the mean of items 2, 7, 9, 14, 15. A lower score thereby conveys a lower perceived level of indifference, overcontrol, or abuse in parental style, and a higher score, vice versa, conveys a higher level of negatively perceived parental style.

### 2.3. Data Analyses

Normality was presumed based on the substantial sample size and subsequently confirmed through visual inspection. All data are given as the mean  $\pm$  standard deviation (SD). Data were analyzed via a t-test and a chi-squared test. To compare the primary outcome variables of the questionnaire scores between ACHD and RC, a linear model adjusted for age and sex was used.

A linear regression model was used to determine whether possible parameters—age, sex, BMI, smoking status, CHD severity, CHD type—and whether CHD underwent surgery or not are associated with total MOPS scores for both mother and father. In the first step, univariable association to MOPS scores was tested for all parameters independently, and in the second step, all significant parameters from the univariable model were combined in a multivariable model. The criterion for a variable to be included in the model was a *p*-value less than or equal to 0.05. Conversely, the criterion for a variable to be removed from the model was a *p*-value greater than 0.10.

All data were analyzed using R Studio (Version 1.2.1335, RStudio, Inc. 2015) with a two-tailed level of significance at *p*-value  $\leq$  0.05.

## 3. Results

After adjusting for age and sex, ACHD reported significantly lower on total MOPS scores regarding both mother (ACHD:  $1.3 \pm 0.01$  vs. RC:  $1.5 \pm 0.03$ ,  $p < 0.001$ ) and father (ACHD:  $1.3 \pm 0.02$  vs. RC:  $1.6 \pm 0.04$ ,  $p < 0.001$ ) and in all three subscales on Indifference (Mother: ACHD:  $1.2 \pm 0.01$  vs. RC:  $1.3 \pm 0.03$ ,  $p < 0.001$ ; Father: ACHD:  $1.3 \pm 0.02$  vs. RC:  $1.7 \pm 0.05$ ,  $p < 0.001$ ), Overcontrol (Mother: ACHD:  $1.6 \pm 0.63$  vs. RC:  $1.9 \pm 0.62$ ,  $p < 0.001$ ; Father: ACHD:  $1.4 \pm 0.52$  vs. RC:  $1.5 \pm 0.50$ ,  $p < 0.001$ ), and Abuse (Mother: ACHD:  $1.2 \pm 0.47$  vs. RC:  $1.4 \pm 0.46$ ,  $p < 0.001$ ; Father: ACHD:  $1.3 \pm 0.59$  vs. RC:  $1.5 \pm 0.57$ ,  $p < 0.001$ ). In other words, ACHD recalled the parental style of both their mother and father as being less indifferent, overcontrolling, and abusive than healthy controls did (Table 2).

In determining the factors that are associated with perceived parental style in ACHD, a univariable linear model revealed several factors to be individually associated with a total MOPS score regarding both the mother (Table 3a) and the father (Table 3b).

Including these significantly associated parameters of the univariable into one multivariable model left female sex ( $\beta = 0.068$ ,  $p = 0.017$ ), higher age ( $\beta = 0.003$ ,  $p = 0.018$ ), and cyanotic CHD ( $\beta = 0.221$ ,  $p = 0.022$ ) to be associated with higher total MOPS regarding the mother. In the multivariable model of the total MOPS score regarding the father, higher age ( $\beta = 0.005$ ,  $p = 0.003$ ), Ebstein anomaly ( $\beta = 0.170$ ,  $p = 0.005$ ), and cyanotic CHD ( $\beta = 0.336$ ,  $p = 0.004$ ) were positively associated. Having never smoked ( $\beta = -0.126$ ,  $p = 0.031$ ) was negatively associated with a higher total MOPS score regarding the father. Put differently, female sex, higher age, Ebstein anomaly, and cyanotic CHD contribute to perceiving the parental style of at least one of the parents negatively.

**Table 2.** Linear model comparing questionnaire scores in CHD vs. RC adjusted for age and sex.

<b>Questionnaire Score Regarding Mother</b>				
	<b>CHD Mean ± SD</b>	<b>RC Mean ± SD</b>	<b>Mean Difference [95% CI]</b>	<b>p-Value *</b>
<b>Total MOPS</b>	1.3 ± 0.01	1.5 ± 0.03	−0.20 [−0.372; −0.142]	<b>&lt;0.001</b>
<b>Indifference</b>	1.2 ± 0.01	1.3 ± 0.03	−0.15 [−0.241; −0.100]	<b>&lt;0.001</b>
<b>Abuse</b>	1.2 ± 0.47	1.4 ± 0.46	−0.22 [−0.296; −0.150]	<b>&lt;0.001</b>
<b>Overcontrol</b>	1.6 ± 0.63	1.9 ± 0.62	−0.26 [−0.357; −0.145]	<b>&lt;0.001</b>
<b>Questionnaire Score Regarding Father</b>				
	<b>CHD Mean ± SD</b>	<b>RC Mean ± SD</b>	<b>Mean Difference [95% CI]</b>	<b>p-Value *</b>
<b>Total MOPS</b>	1.3 ± 0.02	1.6 ± 0.04	−0.26 [−0.341; −0.181]	<b>&lt;0.001</b>
<b>Indifference</b>	1.3 ± 0.02	1.7 ± 0.05	−0.37 [−0.476; −0.267]	<b>&lt;0.001</b>
<b>Abuse</b>	1.3 ± 0.59	1.5 ± 0.57	−0.24 [−0.325; −0.142]	<b>&lt;0.001</b>
<b>Overcontrol</b>	1.4 ± 0.52	1.5 ± 0.50	−0.13 [−0.214; −0.053]	<b>&lt;0.001</b>

\* significant with  $p < 0.05$  are shown in bold, CHD: Congenital Heart Disease, RC: Reference Cohort.

**Table 3.** Parameters associated with the total MOPS score regarding the (a) mother and (b) father.

<b>(a)</b>						
<b>Parameter</b>	<b>Univariable Model Total MOPS Mother</b>			<b>Multivariable Model Total MOPS Mother</b>		
	<b>β</b>	<b>Standard Error</b>	<b>p-Value *</b>	<b>β</b>	<b>Standard Error</b>	<b>p-Value *</b>
<b>Sex (female)</b>	0.075	0.027	<b>=0.005</b>	0.068	0.028	<b>=0.017</b>
<b>Age (years)</b>	0.005	0.001	<b>&lt;0.001</b>	0.003	0.001	<b>=0.018</b>
<b>BMI (kg/m<sup>2</sup>)</b>	0.006	0.003	=0.056			
<b>OP Status (operated)</b>	−0.073	0.034	<b>=0.033</b>	−0.067	0.036	=0.064
<b>Smoking Status</b>						
<b>Non-Smokers</b>	−0.104	0.032	<b>&lt;0.001</b>	−0.064	0.048	=0.184
<b>Ex-Smokers</b>	0.127	0.038	<b>&lt;0.001</b>	0.059	0.057	=0.297
<b>Smokers</b>	0.030	0.048	=0.537			
<b>CHD Severity</b>						
<b>Simple CHD</b>	0.001	0.041	=0.993			
<b>Moderate CHD</b>	0.007	0.028	=0.801			
<b>Complex CHD</b>	−0.007	0.027	=0.805			
<b>CHD Subgroups</b>						
<b>Aortic Stenosis</b>	−0.040	0.037	=0.274			
<b>Tetralogy of Fallot</b>	0.015	0.031	=0.622			
<b>TGA after Switch</b>	−0.090	0.060	=0.139			
<b>TGA §</b>	−0.027	0.057	=0.639			
<b>TGA §§</b>	−0.026	0.044	=0.556			
<b>Isolated Shunts</b>	0.076	0.043	=0.077			
<b>Fontan</b>	0.011	0.060	=0.856			
<b>EBS</b>	−0.016	0.044	=0.722			
<b>Cyanosis</b>	0.225	0.093	<b>=0.016</b>	0.221	0.096	<b>=0.022</b>
<b>CoA</b>	−0.007	0.048	=0.885			

Table 3. Cont.

(b)						
Predictor	Univariable Model Total MOPS Father			Multivariable Model Total MOPS Father		
	$\beta$	Standard Error	<i>p</i> -Value *	$\beta$	Standard Error	<i>p</i> -Value *
Sex (female)	0.067	0.033	<b>=0.040</b>	0.042	0.035	=0.233
Age (years)	0.008	0.002	<b>&lt;0.001</b>	0.005	0.002	<b>=0.003</b>
BMI (kg/m <sup>2</sup> )	0.001	0.004	=0.781			
OP Status (operated)	−0.076	0.041	=0.066			
<b>Smoking Status</b>						
Non-Smokers	−0.133	0.039	<b>&lt;0.001</b>	−0.126	0.058	<b>=0.031</b>
Ex-Smokers	0.129	0.047	<b>&lt;0.001</b>	−0.010	0.069	=0.884
Smokers	0.093	0.059	=0.117			
<b>CHD Severity</b>						
Simple CHD	−0.003	0.049	=0.953			
Moderate CHD	0.041	0.034	=0.228			
Complex CHD	−0.036	0.033	=0.269			
<b>CHD Subgroups</b>						
Aortic Stenosis	−0.048	0.045	=0.286			
Tetralogy of Fallot	−0.024	0.037	=0.516			
TGA after Switch	−0.163	0.072	<b>=0.023</b>	−0.066	0.076	=0.384
TGA <sup>§</sup>	−0.079	0.068	=0.242			
TGA <sup>§§</sup>	−0.010	0.054	=0.846			
Isolated Shunts	0.060	0.052	=0.246			
Fontan	−0.034	0.069	=0.621			
EBS	0.196	0.055	<b>&lt;0.001</b>	0.170	0.060	<b>=0.005</b>
Cyanosis	0.327	0.111	<b>=0.003</b>	0.336	0.116	<b>=0.004</b>
CoA	−0.036	0.057	=0.530			

\* significant with  $p < 0.05$  are shown in bold; CHD: congenital heart disease; <sup>§</sup> Transposition of the Great Arteries after Rastelli and congenitally corrected; <sup>§§</sup> transposition of the great arteries after Senning/Mustared; EBS: Ebstein anomaly; CoA: coarctation of the aorta.

#### 4. Discussion

ACHD recalling parental style as less negative than healthy controls is, on the one hand, surprising considering the large proportion of previously reported psychiatric disorders [18] and emotional distress [8] in complex CHD. Even more so, bearing in mind that 53% of our cohort were of complex severity. Nevertheless, on the other hand, we knew from previous studies that family involvement is very strong in those families, making it less likely that there is high “abuse” and “indifference” in those families. So, this aligns with the general understanding in psychological and sociological research that positive family involvement, communication, and support are associated with healthier child development outcomes. Families that are actively involved in their children’s lives, provide emotional support, and maintain open communication are often linked to better psychological well-being for the children.

To date, this represents the first study evaluating perceived parental style in a large cohort of ACHD. Previous literature on the topic is limited to adolescents, with inconsistent results. Luyckx et al. found no differences in 429 adolescents with CHD compared to 403 healthy controls in perceived parental styles [15]. Cohen and colleagues reported 45 adolescent CHD patients with higher perceived parental acceptance and lower perceived parental control than healthy peers [14].

One factor explaining our findings might be parental overprotection, which has long been known to be an issue in ACHD [19] and is impacting the development of pediatric CHD [20]. While parents of infants and toddlers with CHD appear to experience high levels of parenting stress and poor sleep [21], these parental maladjustments seem to wear off as the children get older [22]. Our results suggest that the parents—as the infants develop into adults—are not necessarily passing on these maladjustments in the form of a

negative parenting style. In fact, it could even be suggested that by engaging more with their chronically ill children, parents are taking especially considerate care of them—the far opposite of indifference or abuse. This argument can be underlined by Brosig et al., who have found a more permissive parenting style in parents of children with hypoplastic left heart syndrome and TGA compared to parents of healthy children [23]. Furthermore, a strong sense of coherence as a coping strategy, which possibly has its foundation in family-related engagement, has been reported to be a prognostic factor for quality of life in CHD patients [3,24,25]. Therefore, in the context of these previous findings, a concern for a notable negative parenting style does not seem to be given based on our results.

In more detail, our analysis found female sex and higher age to be negative contributing factors to recalling parental style of at least one of the parents. These findings are in line with former literature linking emotional distress to female sex [26,27] and older age [2,28]. This underlines the need for targeted interventions in these populations. Especially complex CHD might be at risk for impaired QoL with advanced age [29]. On the other side, a higher age being associated with a more negative parental style could also mean that these patients are older and were therefore young in a time when the handling of bringing up a child with CHD was not as advanced as later on in time. In fact, ACHD aftercare nowadays involves consideration of psychosocial health, and practitioners must be aware of more challenges with a broader scheme of non-cardiac issues than was the case four or five decades ago [30,31]. It is important to approach the interpretation of study findings on parental age with caution, as we cannot definitively determine whether the circumstances have simply changed or if parental age is genuinely a decisive factor.

More than that, we found patients with Ebstein anomaly and cyanotic CHD to also perceive the parental style of at least one of the parents negatively. Patients with cyanosis have, in previous literature, especially been found to be at advanced risk for depression [32], poor academic performance [1], and compromised neuropsychological outcomes [33]. In these patient cohorts as well, it is imperative to acknowledge that the subjects are of higher age, thereby underscoring the potential influence of the conjecture regarding intergenerational disparities as mentioned above. Our findings, along with the above-mentioned literature, support the idea that targeted interventions for specific subgroups at risk have now long been overdue.

Patients with CHD rely on lifelong routine medical care where medical and psychosocial demands may need to go hand in hand. While our findings paint a picture of a less deprecatory parental style in our patients in comparison to healthy controls, they also emphasize the call of previous literature for more psychosocial support in the context of routine medical care in a few herein specified groups at risk. And although this need has been known for a long time, there is currently limited research on the treatment of psychological disorders in CHD patients. Therefore, Jackson and colleagues suggest applying effective forms of treatment to patients with acquired heart disease for new research directions in CHD [8].

#### 4.1. Limitations

Previous literature has suggested that—when it comes to the driving forces behind psychosocial outcomes—not severity classification but rather NYHA class might be a more useful tool in identifying CHD patients at risk [34]. However, almost all of our outpatient patients are in stages I or II, which makes comparison obsolete.

Furthermore, as the influence of sociodemographic factors on psychological health in CHD patients is inconclusive [5], including such data in more detail could have been beneficial for our analysis. However, as generalizations based upon sociodemographic background should generally be avoided, the need for a focus on diagnostic subgroups is provided. Since we used only a questionnaire to assess the reference cohort in our study, it cannot be ruled out that there is a potential selection bias.

#### 4.2. Future Implications

The results challenge previous assumptions and emphasize the need for a differentiated approach to understanding the long-term psychosocial impact of congenital heart defects. One notable future implication is the potential role of parental engagement and support in mitigating the psychological challenges associated with ACHD. This opens avenues for exploring interventions that foster positive parent–child relationships to enhance the well-being of individuals with CHD throughout their lifespan. Additionally, the identified risk factors for recalling negative parental styles, such as female sex and higher age, suggest the importance of targeted interventions for these specific populations. Tailored psychosocial support programs could address the unique needs of female and older ACHD individuals, potentially improving their overall quality of life. Furthermore, the study emphasizes the necessity for ongoing research on psychological disorders in CHD patients, particularly considering the evolving landscape of care over the years. Integrating effective forms of treatment for psychological well-being into routine medical care could be a promising avenue for future investigations, ensuring a comprehensive approach to the holistic care of individuals with CHD.

#### 5. Conclusions

While ACHD recalled the parental style of both their mother and father to be less indifferent, overcontrolling, and abusive than healthy controls did, subgroup analysis revealed specific patients at risk of perceiving the parental style of at least one of the parents negatively. These findings support the need for targeted interventions in specific subgroups susceptible to psychological distress.

**Author Contributions:** J.M. and A.H. were responsible conceptualization, methodology and responsible for data analysis. L.B., J.H. and L.W. sampled the data. L.B. analyzed the data and wrote the first draft. P.E. and R.O.-F. supervised the project. All authors gave important input revising the manuscript. All authors have read and agreed to the published version of the manuscript.

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