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Psychosocial Impact of Congenital Heart Diseases on Patients and Their Families: A Parent's Perspective

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Abstract

Background: Currently, over 90% of children with congenital heart disease (CHD) survive into adulthood. As a consequence the psychosocial impact on children and their families has become an important outcome measure. Therefore, the goal of this study was to assess the psychosocial impact from a parent's perspective and to identify possible predictors. **Methods:** We included all parents of children who underwent open-heart surgery in the years 2010 and 2011 at the Department of Cardiothoracic Surgery at University Hospital Heidelberg and invited them to complete standardized questionnaires. Psychosocial outcome was measured via parent self- and proxy reporting of family burden (Family Burden Questionnaire, FaBel), health-related quality of life (KidScreen-10), developmental problems (Five-to-Fifteen, FTF), and mental health problems (Strength and Difficulties Questionnaire, SDQ). **Results:** In total, 113 families returned the questionnaires completely (71.5%). The Aristotle Basic Complexity score and the STAT 2020 Score overall did not predict the psychosocial impact, whereas the number of surgical operations did significantly predict psychosocial impact across all domains in this study cohort. **Conclusions:** These data suggest that the number of surgical operations might be a relevant predictor for the long-term psychosocial impact on families suffering from CHD and a potential connecting factor for specialized psychological support. When setting up screening instruments or support programs the entire family must be considered.

Keywords

adult congenital heart disease, cardiac (use in combination), cardiology, complications, congenital heart disease (CHD), congenital heart surgery, outcomes (includes mortality, morbidity), quality of life, surgery, complications

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Introduction

Since surgical treatment has been gradually improving over the past decades, the mortality of patients with congenital heart disease (CHD) has significantly decreased.[1] Currently, more than 90% of children with CHD can survive into adulthood.[2] Despite surgical intervention during early childhood, the patients have to cope with the impact of the disease over a lifetime. Concomitantly, the disease affects both the patients' social and familial environment.[3] In addition to the patients' mortality and morbidity, which were the most important outcome measures in the past, quality of life of both children and their families as outcome measure has become the object of past and current research. The results have been highly contradictory.

Several studies reported that the quality of life of children with CHD is poorer compared to healthy children, in particular the influence of motor and cognitive function upon their quality of life.[1,4] In contrast, Reiner et al recently reported a higher quality of life in young patients with CHD than in healthy controls.[5]

Regarding quality of life of the patients' environment, different studies showed that CHD is a source of concern.[6,7] It is not surprising that parents suffer when they accompany their child to hospital for surgical treatment. Landolt et al found that both mothers and fathers reported poorer quality of life compared to population norms at the time of discharge.[8] Yet, after 6 months all mean scores were within or above population norms.

In general, the lack of predictors makes it difficult to identify those who most need specialized support. It has been

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hypothesized that the type of the CHD or the number of operations could be a connecting factor; yet, results have been heterogeneous. Jackson et al reported that families of children with a complex CHD suffer more than families of children with simple cardiac lesions, especially in the domains of familial burden, social relationships, and personal strain.[7] These results were confirmed by other recent studies.[9,10] However, several studies showed no differences between severity classes and diagnostic subgroups.[5,11] Pike et al reported that the number of operations is an independent predictor of memory deficits,[12] and another study suggested that determinants such as distress, hopelessness, or social isolation are more important in explaining reduced quality of life than the severity of the CHD or the number of operations.[6]

All in all, many of the previous studies used a single-measure approach and/or focused on short-term impacts. Therefore, in this study, we investigated the long-term psychosocial impact of CHD on patients and their families, and applied a variety of instruments including family burden, health-related quality of life, mental health problems, and developmental problems. Moreover, most of the previous studies classified the severity of CHD as mild, moderate and complex, even though there is a high diversity in assigning types of CHD to degrees of severity in the literature. Hence, in this study we used the Aristotle Basic Complexity (ABC) score[13] and the STAT 2020 Score[14–16] since they provide a more sophisticated picture than a simple three-part classification. The ABC score is a subjectively derived measure to estimate procedural complexity and has been mainly used as a risk stratification tool to compare quality of care among different hospitals; eg it has been shown that the frequency and severity of complications increases as the technical difficulty increases; but, it has been evaluated in quality of life studies as well.[11] The STAT 2020 Score is lacking with respect to morbidity and technical difficulty components of the ABC score but is empirically derived and reflects the statistically estimated risk of mortality.[16]

The purpose of the present study was firstly to investigate the psychosocial impact of CHD among our study population, and secondly to determine possible long-term predictors. It was hypothesized that (i) parents of children with higher ABC or (ii) STAT 2020 Score would report experiencing a more negative impact of CHD than parents of children with lower scores, and (iii) that parents of children with a higher number of operations would report experiencing a higher psychosocial impact than parents of children with a lower number.

Patients and Methods

Participants

This prospective study was performed to evaluate the long-term psychosocial impact (ie family burden, health-related quality of life, developmental problems, and mental health problems) of both children suffering from CHD and their families. The study was approved by the Ethics Committee of the Medical Faculty at the University of Heidelberg (S-044/2017). The study focused on a consecutive sample of all families whose children underwent surgical treatment at the Department of Cardiothoracic Surgery at University Hospital Heidelberg in the years 2010 and 2011. Data was collected between April and October 2019. All families were recruited after the surgical procedure. Informed consent was obtained from all participants.

Procedure

A total of 349 affected families were called to obtain a current email address. Excluded were those families who (a) were not available ($N = 69$), (b) did not wish to participate ($N = 14$), (c) did not speak proper German ($N = 12$), (d) lived abroad ($N = 6$), and (e) the families whose children either passed away or were older than 18 at time of recruitment ($N = 90$). Thus, a total of 158 families were enrolled and received a link to access a secure online survey tool. All questions were answered by the parents since no self-reporting was possible in most cases due to the young age of the children. Informed consent was obtained from all participating families.

Measures

Parents were asked to complete a compilation of general questions, such as gender, age, size, and weight of their child as well as number of operations and date of last surgical operation. These questions were followed by a set of questionnaires, which addressed the fact that the psychosocial status is multifactorial. Therefore, the set included one questionnaire about the family itself (Family Burden Questionnaire, FaBel) and three about the child's situation in particular (KidScreen-10, Five-to-Fifteen (FTF), Strength and Difficulties Questionnaire (SDQ)). Additional information can be found in the Supplement.

Statistics

Three main variables were investigated as predictors of child and family outcome: (i) the ABC score,[13] (ii) the STAT 2020 Score[14–16], and (iii) the number of conducted operations.

The ABC score was first published in 2004 by a group of 50 pediatric surgeons from 23 countries as a new method of evaluation of quality of care in CHD based on the complexity of the surgical procedure. It is a consensus of expert opinion and was called Aristotle in reference to the philosophy of Aristotle "Where there is no scientific answer available, the opinion perceived and admitted by the majority has value of truth" (Rhetoric, Book I, 350 BC). The ABC score is based on three factors: the potential for operative mortality, the potential for operative morbidity, and the anticipated technical difficulty. Each surgical procedure received a score for each of the three factors ranging from 0.5 to 5.0, forming a score which ranged from 1.5 to 15. If surgical therapy included multiple operations, the highest score was taken. The original STAT Score was first published in 2009[14] and has been updated in 2020.[16] It was developed as an objective, empirically based index to identify the statistically estimated risk of in-hospital mortality. Each

procedure was assigned a numeric score ranging from 0.1 to 5.0 based on the estimated mortality rate.

Pearson's correlations were conducted to assess associations among the psychosocial impact on the one hand, and ABC score, STAT 2020 Score, and number of operations on the other hand.

In addition, for each of the outcome variables linear regression models were calculated: one with the ABC score, STAT 2020 Score and number of operations as predictors, and one with the addition of the covariates gender, age, size-per-age percentiles, weight-per-age percentiles, and time since last operation. For each of the outcome variables we compared the model with the covariates and the model without the covariates with a likelihood ratio test to verify that the relation between outcome and predictors is not confounded by covariates.

A value of $P < .05$ was considered significant. All analyses were performed with the statistical software Stata 15.

Results

Of the 158 families who were invited to participate, 113 returned the set of questionnaires completely (71.5%). Detailed characteristics are displayed in Table 1. The various operations that were conducted are listed in Table 2. The most frequent types of CHD were atrial septal defect, ventricular septal defect, patent ductus arteriosus, coarctation of the aorta, tetralogy of Fallot, and transposition of the great arteries.

Relation Between Psychosocial Impact and the Complexity of the Surgical Procedure or the Estimated Mortality Risk of the Disease

Hypothesis 1 and 2 predicted that parents of children with higher ABC or STAT 2020 Score, respectively, would experience a more negative psychosocial impact of CHD than parents of children with lower scores.

Table 1. Study Characteristics.

	N (%) or mean ± SD (range)
Total study population	113
Parent answering the questionnaire	
Father	34 (30.09%)
Mother	79 (69.91%)
CHD-affected children	
Age (years)	8.17 ± 3.40 (5-17)
Weight (kg)	27.60 ± 14.26 (15-90)
Percentile weight	25.44 ± 27.60 (1-95)
Height (cm)	128.06 ± 19.80 (90-182)
Percentile height	32.97 ± 30.41 (1-99)
Number of operations	2.15 ± 1.58 (1-10)
Time since last operation (years)	5.07 ± 2.17 (0.06-7.90)
ABC score	6.2 ± 2.2 (3.0-11.0)
STAT 2020 Score	0.4 ± 0.4 (0.1-1.6)

Abbreviations: CHD, Congenital Heart Disease; ABC, Aristotle Basic Complexity.

However, there were no significant associations between the ABC score on the one hand and the measures FaBel, KidScreen-10, FTF, and SDQ on the other hand, except for FaBel's subcale 'personal strain' and FTF's subscales 'gross motor skills' and 'memory' (Table 3). Similar results were found when the STAT 2020 Score was used. Only the KidScreen-10 as well as the total burden and the two subscales 'personal strain' and 'concern for siblings' of FaBel showed significant correlations.

Relation Between Psychosocial Impact and the Number of Operations

Hypothesis 3 predicted that parents of children with a higher number of surgical operations would report experiencing a higher psychosocial impact than parents of children with a lower number, and indeed the number of performed surgical procedures significantly predicted negative psychosocial impact of CHD across all investigated domains (Table 3). Notably, there was no association between the two measures, number of operations and highest ABC score (Supplemental Table 1).

Regression Analyses

Regression analyses were performed with and without the covariates gender, age, size-per-age percentiles, weight-per-age percentiles, and time since last operation. For all regression models, a likelihood test indicated that the unadjusted model had the better fit. Therefore, only the unadjusted model is presented in Table 4.

Discussion

The results of this study suggest that regarding the familial situation, the complexity of the surgical procedure, represented by the ABC score, and the estimated mortality risk of the CHD, represented by the STAT 2020 Score, are overall not significantly associated with the long-term psychosocial impact. Only a limited number of subscales were found to be significant. In contrast, the number of surgical operations significantly predicted the negative psychosocial impact of CHD on the familial environment over 5 years after surgery.

These findings are in line with comparable older[3,17] and newer studies,[18] even though the measures and nature of cardiac abnormalities differed. For example Vrijmoet-Wiersma et al used the 15-item self-reporting questionnaire *Pediatric Inventory for Parents* and found that parents of children with hypoplastic left heart syndrome reported a more negative impact of CHD on family functioning than parents of children with transposition of the great arteries.[19] Since hypopolastic left heart syndrome and transposition of the great arteries are both considered as severe CHD, the major discriminating factor is the number of operations. Notably, some studies suggest that caring for a child with hypoplastic left

Table 2. Performed Surgical Procedures Ascending in Aristotle Basic Complexity Score.

Procedure	ABC score	STAT 2020 Score	Average number of operations
Atrial Septal Defect (ASD) repair; *N* = 12	3.0	0.1	1.5
Patent Foramen Ovale (PFO), primary closure; *N* = 2	3.0	0.1	1.0
Pacemaker implantation, permanent; *N* = 7	3.0	0.2	5.1
Patent Ductus Arteriosus (PDA) closure, surgical; *N* = 7	3.0	0.3	3.0
Shunt, ligation and takedown; *N* = 1	3.5	0.3	2.0
Partial Anomalous Pulmonary Venous Connection (PAPVC) repair; *N* = 1	5.0	0.1	1.0
Ventricular Septal Defect (VSD) repair; *N* = 12	6.0	0.1	1.2
Coarctation repair			
End to end; N = 7	6.0	0.2	1.4
Patch aortoplasty; N = 1	6.0	0.2	1.0
End to end, extended; N = 1	8.0	0.2	1.0
Vascular ring repair; *N* = 3	6.0	0.1	1.0
Pulmonary Artery Banding; *N* = 5	6.0	1.2	3.2
Pulmonary Artery Stenosis			
Reconstruction (plasty), main (trunk); N = 1	6.0	0.2	5.0
Reconstruction (plasty), branch, periphery (at or beyond hilar bifurcation); N = 2	7.8	0.3	2.5
Aortic Stenosis repair			
Subvalvar; N = 1	6.3	0.2	2.0
Supravalvar; N = 1	7.5	0.4	1.0
Shunt, systemic to pulmonary, Modified Blalock–Taussig Shunt (MBTS); *N* = 1	6.3	0.9	4.0
Right Ventricular Outflow Tract Obstruction procedure; *N* = 3	6.5	0.2	2.3
Bidirectional cavopulmonary anastomosis (bidirectional Glenn); *N* = 2	6.8	0.3	2.5
Aortic arch repair; *N* = 5	7.0	0.5	1.6
Valvuloplasty			
Tricuspid; N = 1	7.0	0.2	1.0
Mitral; N = 1	8.0	0.3	2.0
Aortic; N = 4	8.0	0.2	1.0
Valve replacement			
Tricuspid; N = 1	7.5	0.7	3.0
Mitral; N = 1	7.5	0.6	1.0
Conduit			
Placement, right ventricle to pulmonary artery; N = 5	7.5	1.6	2.8
Reoperation; N = 3	8.0	0.1	2.7
Tetralogy of Fallot (TOF) repair			
Ventriculotomy, nontransannular patch; N = 1	7.5	0.1	4.0
Ventriculotomy, transannular patch; N = 2	8.0	0.2	1.5
No ventriculotomy; N = 8	8.0	0.1	2.6
Aortic aneurysm repair; *N* = 1	8.8	0.2	1.0
Rastelli; *N* = 1	10.0	0.2	2.0
Arterial switch operation; *N* = 5	10.0	0.4	1.4
Anomalous origin of coronary artery from pulmonary artery repair; *N* = 1	10.0	0.3	4.0
Double Outlet Right Ventricle (DORV), intraventricular tunnel repair; *N* = 1	10.3	0.5	2.0
Konno procedure; *N* = 1	11.0	0.4	1.0
Pulmonary atresia repair; *N* = 1	11.0	1.3	2.0

Abbreviations: ABC, Aristotle Basic Complexity.

heart syndrome is more stressful than caring for a child with other chronic illnesses, such as inflammatory bowel disease, cancer, diabetes or sickle cell disease.[20,21]

With regard to the child's long-term outcome, we found that a higher number of operations is associated with an impaired health-related quality of life, delayed state of development, and emotional and behavioral problems (mental health problems). The complexity of the surgical correction was only associated with impaired 'gross motor skills' and 'memory' and the statistically estimated risk of mortality showed no significant correlation.

These results are consistent with some studies,[5,22,23] while others suggest on the one hand that more than one cardiac surgical intervention makes no difference with respect to the quality of life in adulthood[24] and on the other hand that the severity of the CHD does appear to have an impact on the child's quality of life.[9,10] These discrepancies are difficult to explain.

In general, it should be taken into account that most quality-of-life studies with patients with CHD use different questionnaires. Furthermore, there is a major difference between self-reported and parent proxy-reported study design – parents tend to report a lower health-related quality of life

Table 3. Summary of Pearson's Correlations.

	Number of operations			ABC score			STAT 2020 Score		
	R	*P*	95% CI	R	*P*	95% CI	r	*P*	95% CI
FaBel									
Daily social impact	−0.354	<.001***	[−0.51, 0.18]	−0.063	.511	[−0.24, 0.12]	−0.180	.057	[−0.35, 0.01]
Personal strain	−0.337	<.001***	[−0.49, −0.16]	−0.214	.023*	[−0.38, −0.03]	−0.281	.003**	[−0.44, −0.10]
Financial impact	−0.372	<.001***	[−0.52, −0.20]	−0.089	.347	[−0.27, 0.10]	−0.137	.148	[−0.31, 0.05]
Concern for siblings	−0.307	.003***	[−0.48, −0.11]	−0.161	.128	[−0.36, 0.05]	−0.247	.019*	[−0.43, −0.04]
Problems in coping	−0.124	.191	[−0.30, 0.06]	0.051	.593	[−0.14, 0.23]	−0.062	.516	[−0.24, 0.12]
Total burden	−0.390	<.001***	[−0.54, −0.22]	−0.109	.251	[−0.29, 0.08]	−0.235	.012*	[−0.40, −0.05]
KidScreen-10	−0.393	<.001***	[−0.54, −0.22]	−0.016	.868	[−0.20, 0.26]	−0.224	.017*	[−0.39, 0.04]
FTF									
Gross motor skills	0.299	.001***	[0.12, 0.46]	−0.193	.040*	[−0.36, −0.01]	0.016	.869	[−0.17, 0.20]
Fine motor skills	0.243	.010**	[0.06, 0.41]	−0.052	.586	[−0.23, 0.13]	−0.071	.457	[−0.25, 0.12]
Memory	0.318	<.001***	[0.14, 0.47]	−0.232	.013*	[−0.40, −0.05]	−0.008	.930	[−0.19, 0.18]
SDQ									
Emotional problems	0.133	.160	[−0.05, 0.31]	−0.029	.757	[−0.21, 0.16]	0.180	.056	[−0.01, 0.35]
Conduct problems	0.165	.081	[−0.02, 0.34]	−0.060	.527	[−0.24, 0.13]	0.027	.778	[−0.16, 0.21]
Hyperactivity/inattention	0.258	.006***	[0.08, 0.42]	−0.118	.215	[−0.30, 0.07]	0.033	.728	[−0.15, 0.22]
Peer relationship problems	0.267	.004***	[0.09, 0.43]	−0.113	.235	[−0.29, 0.07]	0.064	.501	[−0.12, 0.25]
Prosocial behavior	−0.255	.007***	[−0.42, −0.07]	0.021	.826	[−0.16, 0.20]	−0.049	.608	[−0.23, 0.14]
Total problem	0.272	.004***	[0.09, 0.44]	−0.107	.257	[−0.29, 0.08]	0.098	.303	[−0.09, 0.28]

Abbreviations: ABC, Aristotle Basic Complexity; FaBel, Family Burden Questionnaire; FTF, Five-to-Fifteen; SDQ, Strength and Difficulties Questionnaire.
*$P < .05$, **$P < .01$, ***$P < .001$.

Table 4. Regression Analyses.

	M1 (unadjusted)		
	Number of operations	ABC score Coefficient [95% CI]	STAT 2020 Score
FaBel (total)	−0.127*** [−0.179, −0.074]	−0.038* [−0.075, −0.001]	−0.195* [−0.381, −0.010]
KidScreen-10	−1.638*** [−2.346, −0.931]	−0.231 [−0.732, 0.270]	−2.434 [−4.911, 0.043]
SDQ (total)	1.242** [0.370, 2.114]	−0.220 [−0.838, 0.397]	−0.994 [−2.100, 4.088]
FTF			
Gross motor skills	0.103** [0.036, 0.171]	−0.040 [−0.088, 0.008]	−0.039 [−0.281, 0.204]
Fine motor skills	0.076* [0.017, 0.135]	−0.003 [−0.044, 0.039]	−0.122 [−0.330, 0.085]
Memory	0.097** [0.037, 0.157]	−0.044* [−0.087, −0.002]	−0.067 [−0.283, −0.149]

Abbreviations: ABC, Aristotle Basic Complexity; FaBel, Family Burden Questionnaire; FTF, Five-to-Fifteen; SDQ, Strength and Difficulties Questionnaire.
*$P < .05$, **$P < .01$, ***$P < .001$.

of their child,[25,26] which may refer to the so-called "vulnerable child syndrome".[21] Moreover, the age seems to be of importance. Older patients with CHD tend to report a lower quality of life,[5] as do healthy peers all across Europe,[27] so that the question remains whether there is a significantly larger decline in quality of life in patients with CHD. Interestingly, the intercountry variation in illness perceptions seems to be generally small.[28]

While it has been previously suggested that the number of prior cardiac operations does not seem to correlate with the patients' reported quality of life,[24] this might be most likely be explained by a different study design and patient sample. In contrast to our approach, Wang et al examined adults with CHD via self-reporting. Other factors to be considered are the disability paradox[29] or the sense of coherence.[30]

The strengths of this study included the large number of families enrolled and a high response rate. Moreover, the study compared a broad variety of CHD diagnoses, which makes this approach more generalizable versus a limited comparison. An additional strength is our web-based data collection system. Web-based assessment has several advantages: decreased experimenter demand and social desirability effects, reduced missing data, avoidance of data entry errors, savings of money and time, and possibly greater self-disclosure by participants.[31]

Limitations

Despite these interesting results, there are a few study limitations to consider. Firstly, the data included solely parent self- and proxy report. Older patients' self-report may potentially have provided additional information, as has been suggested by Menahem et al[32,33] Secondly, we had an overrepresentation of mothers answering the questionnaires, so that findings may not be generalized. Thirdly, the observed associations could be accounted for by the existence of confounders, such as surgical outcome or further surgical interventions that were not available for analysis. Fourthly, the study cohort has a small number of patients with functionally univentricular heart disease, so that in these more complex CHD patients the results may differ. Lastly, since the last surgical operation took place years ago in most cases, there is a risk of overreporting bias: We cannot predict whether participation was dependent on psychosocial burden and, for example, parents with a higher burden may have been more apt to respond.[19] Yet, as suggested by Spijkerboer et al,[34] the majority of nonresponders could have also been parents with posttraumatic stress symptoms, who were unable to revisit the surgical operation leading to an underrepresentation of psychosocial burden.

Conclusions

In conclusion, the present study shows that the number of operations might be a valuable predictor for the long-term psychosocial impact of CHD on patients and their families. Building on this foundation, the number of operations is a promising connecting factor, which may be used to identify and provide support for families at risk. Even in a setting of limited resources, sufficient financial means are necessary to enable families to cope with the sometimes laborious surgical treatment of CHD. Based upon these results, we think it is prudent to be open to the use of reliable and valid questionnaires as screening instruments for high psychosocial stress experienced by the parents and their affected children in routine follow-ups. Ideally, specific and easy-to-use questionnaires would be developed for this purpose by a multidisciplinary team. Furthermore, future research should concentrate on evaluating the appropriate strategy for specialized psychosocial support since long-term effects are becoming apparent.

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Supplemental Material

Supplemental material for this article is available online.

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