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Psychosocial burden and quality of life of parents with children with univentricular hearts compared to ASD parents and parents of heart-healthy children

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Abstract

Background: Patients with univentricular hearts can only be palliated by a staged surgical procedure that carries a high morbidity and mortality risk. The aim of this study was to examine the emotional demands, psychosocial burden, and quality of life of parents with children with univentricular hearts compared to parents of children with a simple heart defect, those with no heart defect and children with chronic diseases. *Methods:* An anonymous questionnaire was created to interview parents about their quality of life, stressors, needs, strategies for coping with illness, and partnership satisfaction. *Results:* 73 families participated in the study. Parents of children with univentricular hearts experience a significantly higher psychosocial burden, limitations in daily life, and distress in family interactions, as well as greater emotional distress compared to the other study groups. When comparing the families of children with other chronic diseases (e.g. cystic fibrosis, chronic arthritis and diabetes), these differences remained significant. *Conclusion:* The study confirms a higher psychosocial burden, restrictions in daily life and a lower quality of life of parents with children with univentricular hearts, compared to parents of children with simple heart defects and parents of heart-healthy children or those with other chronic diseases. Since this condition persists until adolescence and adulthood, the families are exposed to special challenges and stresses throughout their lives. This has yet to be adequately addressed in the management of these families.

Introduction

Children with univentricular hearts are considered to be the most seriously affected group among children with congenital heart defects (CHDs). Because no true anatomical correction of the CHD is possible, only palliation/improvement and haemodynamics remain unphysiological throughout life (univentricular palliation).

Even though most patients in countries like Germany, where the operation is performed, are now able to reach adulthood, those affected require lifelong medical care and face a lifetime of limitations.[1]

This highly complex group of patients has a particular medical history, usually involving numerous surgeries, re-operations, and catheter examinations. This leads to significant physical, neurological and psychosocial morbidity, and an increased risk of mortality. Corrective surgeries are usually performed with cardiopulmonary bypass in the neonatal and infant period, but even afterwards a significantly increased risk of developing psychomotor and psychosocial developmental disorders in the long-term course remains.[2–4] In addition, a moderately severe degree of psychological impairment and other chronic diseases are often seen in these children.[5] The prevalence of neuropsychological disorders is much higher in univentricular heart children with 65% compared to 22% in the reference population.[6]

Besides medical aspects, psychosocial problems and stress play an important role for the affected children and their parents and, if present, their siblings.[4] Because of this, the parents of these children may face extraordinary challenges both as individuals, as couples and within their partnership. The same applies to the siblings, whose mental health and quality of life can be similarly affected.[7]

There are now several studies that address the consequences of the disease for the children; studies however that focus on the parents, and here in particular on their relationship, are scarce.[8] Rempel et al. used a qualitative approach to examine the different phases of pressure on parents of children with hypoplastic left heart syndrome.[9] Our study wants to take a quantitative approach to the demands and stresses faced by parents of a child with univentricular heart. In particular, the focus lies on the experience of the parents as individuals in relation to the disease of their child and its effects on the satisfaction with the partnership, the stability of the partnership, and the situation of the siblings. Thus, the aim of this work was to identify problem

areas and gaps in coverage to offer potential solutions for better care of the parents or families of these children.

Methods

Study design and participants

In this monocentric, cross-sectional survey study, participants were recruited from the database of the outpatient clinic and inpatient ward of the Department of Pediatric Cardiology and Pediatric Intensive Care Medicine at the LMU Klinikum, Munich. All families with a child born with an anatomical or functional univentricular heart defect who underwent Fontan completion were included. Families with a child with a simple heart defect, in our study atrial septal defect (ASD), were recruited from patients who underwent ASD correction at the Department during the years 2016 to 2019. The control group of families in which there is no child with a heart defect were recruited through schools and colleagues. Parents with insufficient knowledge of the German language were not included to allow completion of the questionnaire without assistance. The participants received a nine-page paper questionnaire for each parent comprising four survey instruments (see below). The completed questionnaires were returned anonymously by the parents for questionnaire analysis. Single parents only returned one questionnaire. The study was approved by the ethics committee of the LMU Munich on the 13.06.2017 and conducted in accordance with the revised version of the Declaration of Helsinki.

Questionnaires

Four individual survey instruments were used: "The Family Stress Questionnaire," "The Ulm Quality of Life Inventory for Parents of Chronically Ill Children," "The Partnership Questionnaire," and the "Needs Scale for Parents of Chronically Ill Children".[10–13]

The survey and evaluation focused on the areas of quality of life, burden of disease, and coping with the disease. Furthermore, the requirements for additional support in the private and social sphere were investigated. Lastly, the quality and stability of the parental partnership were explored.

The survey instrument on the psychosocial burden of disease was the German version of "The family Stress Questionnaire" (FaBel), which corresponds to "The Impact on Family Scale Instrument," and is widely used in Anglo-American countries.[10] It consists of 33 items on a four-point rating scale from "does not apply at all" to "largely applies". As the questionnaire refers to parents of an ill child, only the two study groups, parents of a child with univentricular heart and ASD, were compared to each other.

Several well-established instruments for the assessment of the quality of life of children with CHD exist.[14] Special instruments that address the quality of life of parents with chronically ill children are less common. The quality of life survey instrument, "The Ulm Quality of Life Inventory for Parents of Chronically Ill Children" (ULQIE), is a survey instrument that measures the quality of life of parents of chronically ill children during the last week (in the last 7 days).[15] Quality of life can be defined both negatively, in terms of the absence of distress, and positively, in terms of well-being, performance, and satisfaction. The questionnaire includes 29 items with five subscales, some items referring directly to the child's illness. 19 items are formulated positively and thus include aspects of well-being, satisfaction, and functioning. 10 items are phrased negatively and cover complaints or subjectively perceived burdens that represent limitations in the patients' parents' quality of life. Questions are answered on a five-level scale (0 = "never," 1 = "rarely," 2 = "sometimes," 3 = "often," 4 = "always"). The quality of life assessment refers to the last seven days in to make short-term changes in quality of life detectable.[11] To obtain a broader study group, parents of heart-healthy children and parents of children with univentricular heart and ASD were included.

The partnership survey instrument, "The partnership questionnaire" (PFB) was developed to assess partnership quality.[12] It consists of 29 items belonging to three scales: dispute behaviour, tenderness, commonality/communication, plus an item for a global happiness assessment. Response scales are four-level (0 = "never/very rarely," 1 = "rarely," 2 = "often," 3 = "very often"). The global happiness assessment is six-level to assess how happy respondents are with their partnership (0 = "very unhappy" to 5 = "very happy"). Item two of the PFB questionnaire was excluded in this study, because of the intimate nature of the question and an expected negative impact on the response rate. For this questionnaire, parents with heart-healthy children were included, as the questionnaire was not specific to the experience of illness.

To assess the needs of the parents, the survey instrument of the German "Bedürfnis-Skala für Eltern chronisch kranker Kinder" was applied. This instrument was developed by Wiedebusch and Muthny to examine the needs of parents of chronically ill children.[16] The "Needs Scale for Parents of Chronically Ill Children" has 19 items on a five-point rating scale from "not at all" to "very strongly". Parents can rate their need for further information concerning the disease, treatment, and dealing with authorities. Results were compared to published data of parents and families of children with haemophilia, diabetes mellitus type 1, and juvenile idiopathic arthritis.[16] As the questionnaire is specifically to the needs of parents of chronically ill children, only two study groups, parents of a child with univentricular heart and ASD, were compared with each other.

Statistical analysis

The paper survey answers were transferred to an Excel spreadsheet. Statistical data analysis and evaluation were performed with IBM SPSS Statistics for Windows, Version 26.0 (Armonk, NY: IBM Corp.). The responses to the global happiness scale of the PFB were presented descriptively in a frequency table. For this purpose, the frequency and percentage of the highest agreement ("often" and "very often") among all participants was calculated.

Participants' characteristics are presented as the number of participants and percentages for categorical variables and median and interquartile ranges for continuous variables. Characteristics of the study groups were compared by Kruskal Wallis Test, Mann–Whitney Test or Chi-squared test and Fisher's exact test. Employment status was only tested between the full-time and part-time groups, since the groups "unemployed" and "others" were very small. The parent's marital status was also compared between "married" and all others groups combined, because "relationship with partner," "divorced," "widowed" and "other" were very scarce. Demographic data specific to children with heart defects was only tested for children in the parents of a child with univentricular heart and the ASD group.

Mean values were calculated for all subscales of each questionnaire and compared by group using the t-test. The significance level was set at $p = 0.05$. Parents of univentricular

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Table 1. Patients' characteristics

		Study group			
	Total n (%) or median ± SD	Parents of a child with univentricular heart n (%) or median ± SD	ASD n (%) or median ± SD	Heart healthy n (%) or median ± SD	p-value
Number of patients (%)	73 (100)	49 (67.1)	13 (17.8)	11 (15.1)	–
Age of parent (years)	44.74 ± 6.94	44.49 ± 7.13	41.62 ± 6.17	49.55 ± 4.39	0.140
Sex of parent (female)	39 (53.4)	26 (53.1)	7 (53.8)	6 (54.5)	0.996
Employment status					0.549
Unemployed	3 (4.1)	3 (6.1)	–	–	
Full-time	35 (47.9)	25 (51.0)	6 (46.2)	4 (36.4)	
Part-time	26 (35.6)	15 (30.6)	6 (46.2)	5 (45.5)	
Other	9 (12.3)	6 (12.2)	1 (7.7)	2 (18.2)	
Parent's marital status					0.886
Married	63 (86.3)	42 (85.7)	11 (84.6)	10 (90.9)	
Relationship with partner	6 (8.2)	6 (12.2)	–	–	
Divorced	1 (1.4)	–	–	1 (9.1)	
Widowed	1 (1.4)	–	1 (7.7)	–	
Other	2 (2.7)	1 (2.0)	1 (7.7)	–	
Number of children in the household	1.9 ± 0.80	2.0 ± 0.87	1.8 ± 0.38	1.6 ± 0.84	0.700
Age of child with heart defect	11.3 ± 6.94	12.2 ± 7.42	7.6 ± 2.43	–	<0.001
Sex of child with heart defect (female)	24 (38.7)	18 (36.7)	6 (46.2)	–	0.7645
Parent's perceived severity of child's disease					
Healthy	13 (21.0)	–	13 (100)	–	
Mild	9 (14.5)	9 (18.4)	–	–	
Moderate	17 (27.4)	17 (34.7)	–	–	
Severe	16 (25.8)	16 (32.7)	–	–	
Very severe	6 (9.7)	6 (12.2)	–	–	

heart children were compared with two control groups. The first control group comprised families with a child with ASD, and the second control group included families with heart-healthy children. Due to the small sample size of the heart-healthy group, only the parents of a child with univentricular heart and the ASD group were included in the t-tests. To check the internal consistency of the questionnaires, Cronbach's alpha was calculated for all subscales.

Results

The survey was conducted from summer of 2017 to summer of 2019. A total number of 387 parents, recruited either from the outpatient clinic database or directly from the inpatient ward, were contacted. Interest in participating in the study was expressed by 217 parents, who then received the questionnaire by mail or handed out in person. Among them were 134 parents with a child with univentricular heart, 47 with a child with ASD, and 38 parents whose children did not have a heart defect. Final participation consisted of 73 parents. Of these, 49 had a child with univentricular heart, 13 had a child with an ASD, and 11 had children without a heart defect. Details of the patients and families are presented in Table 1.

Psychosocial stress

Since the primary focus was on the psychosocial stress of the parents caused by the child's illness, only the two study groups, parents of a child with univentricular heart and ASD, were compared.

The assessment of the "daily social load" was significantly different between ASD and univentricular heart parents (univentricular heart: MV = 1.93, SD = 0.489, ASD: MV = 1.24, SD = 0.203, p-value < 0.001). A similar difference was found for the "financial charges" with a significant difference of 0.68 for the mean values of the parents of a child with univentricular heart (MV = 1.83, SD = 0.662) and the ASD (MV = 1.15, SD = 0.298) group (p-value < 0.001). When assessing the item "stress on siblings," parents of a child with univentricular heart (MV = 1.79, SD = 0.801) also showed a statistically significant difference of 0.7 compared to the ASD parents (MV = 1.09, SD = 0.216, p-value < 0.001). In the area of "personal burden and worries about the future" the parents of a child with univentricular heart showed a

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Table 2. Comparison of mean values of all subscales and study groups on the psychosocial stress of parents and the parental quality of life

	Study group						
	Parents of a child with univentricular heart		ASD		Heart-healthy		
Subscales	N	Mean value (SD)	N	Mean value (SD)	N	Mean value (SD)	p-value
Psychosocial stress of parents							
Daily social load	49	1.93 (0.489)	13	1.24 (0.203)			<0.001[a]
Personal burden and worries about the future	49	2.38 (0.614)	13	1.25 (0.484)			<0.001[a]
Financial charges	49	1.83 (0.662)	13	1.15 (0.298)			<0.001[a]
Stress siblings	41	1.79 (0.801)	11	1.09 (0.216)			<0.001[a]
Coping problems	49	0.72 (0.647)	13	0.95 (0.718)			0.3139[a]
Parental quality of life							
Physical / daily functioning	47	2.62 (0.665)	13	3.20 (0.838)	11	3.19 (0.492)	0.0363[b]
Satisfaction with family	47	3.04 (0.755)	13	3.22 (0.808)	11	3.39 (0.474)	0.4939[b]
Emotional stability	47	2.42 (0.929)	13	3.38 (0.747)	11	2.89 (0.610)	<0.001[b]
Well-being	47	2.69 (0.718)	13	3.29 (0.828)	11	3.02 (0.569)	0.0295[b]
Self-development	47	1.92 (0.994)	13	2.50 (1,031)	11	2.66 (0.422)	0.0858[b]
ULQIE—Overall scale	47	2.57 (0.658)	13	3.15 (0.727)	11	3.10 (0.333)	0.0190[b]

[a]The p-value reflects the comparison between parents of a child with univentricular heart and ASD.
[b]The p-value reflects the comparison between parents of a child with univentricular heart, ASD, and heart-healthy.

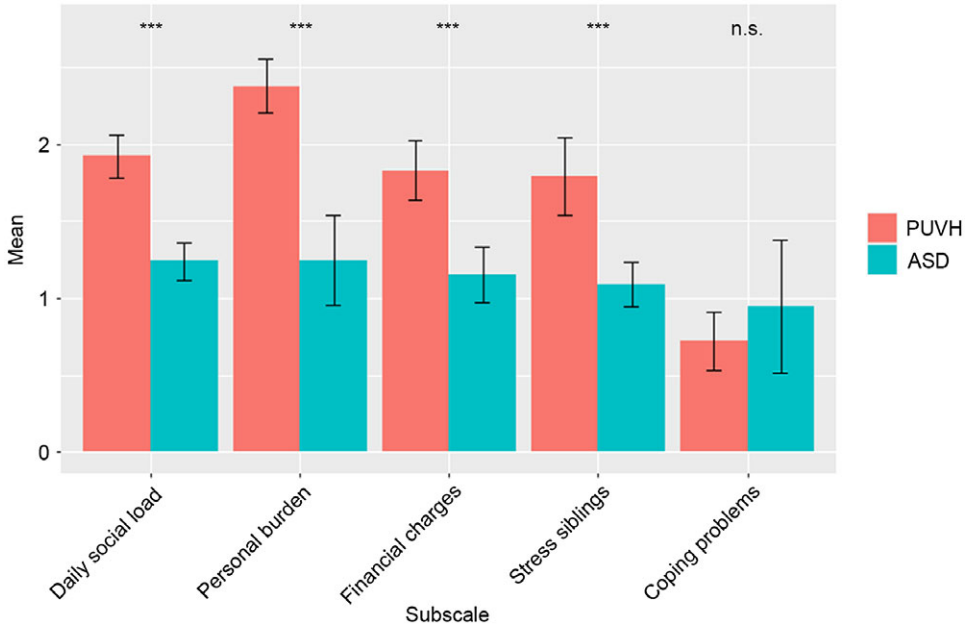

Figure 1. Psychosocial stress in families with children with disabilities.

noticeably higher value (MV = 2.3, SD = 0.614) than the ASD (MV = 1.25, SD = 0.484) with a mean difference of 1.13 (p-value < 0.001). The univentricular heart parents seem to be exposed to greater personal stress or future worries than those in the ASD group. See Figure 1 and Table 2 for a graphical representation of the subscales of the FaBel questionnaire.

Partnership satisfaction

Regarding partnership satisfaction, a clear trend but no significant difference could be identified between the three groups, which lay in a lower partnership satisfaction of the univentricular heart parents (54%) when compared to the ASD and heart-healthy parents (60%). ASD parents displayed the lowest "dispute"

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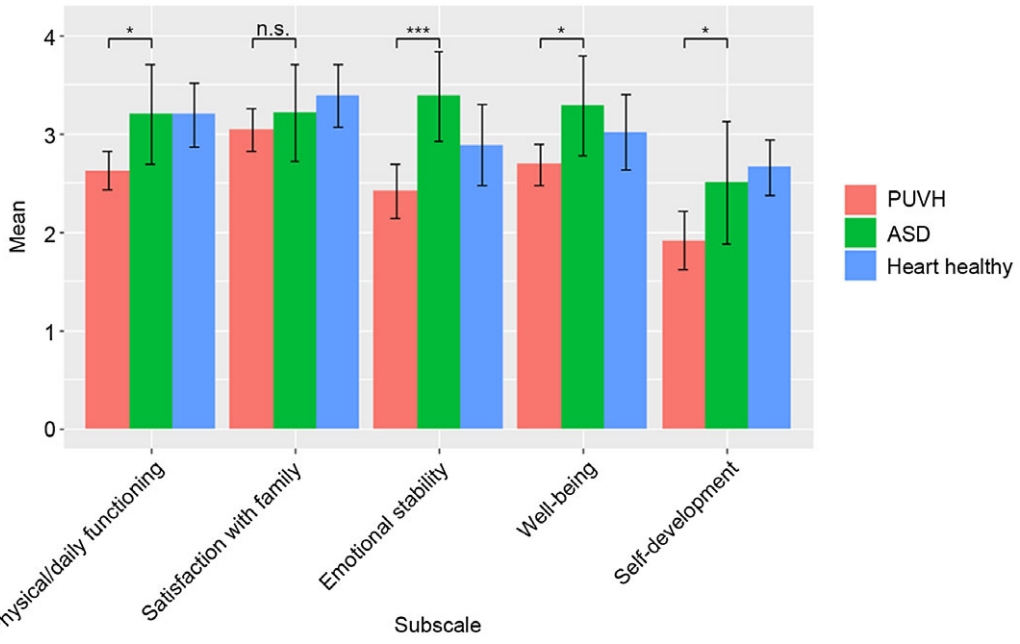

Figure 2. Parental quality of life.

behaviour (MV = 0.67, SD = 0.727), followed by parents of a child with univentricular heart (MV = 0.70, SD = 0.552) and heart-healthy (MV = 0.71, SD = 0.570). ASD parents also had the highest "tenderness" values (MV = 1.76, SD = 0.869), followed by very similar results for the heart-healthy parents (MV = 1.70, SD = 0.606) and non-significant but markedly lower values for the parents of a child with univentricular heart (MV = 1.39, SD = 0.634). "Communication/communality" was also slightly lower in the parents of a child with univentricular heart group (MV = 1.80, SD = 0.513), compared to ASD (MV = 1.99, SD = 0.803) and heart-healthy parents (MV = 2.03, SD = 0.707). Statistical significance was not reached for any of the comparisons, but there are indicators that tenderness and communication are slightly impaired in the parents of a child with univentricular heart group, possibly due to the high demands of caring for a chronically ill child.

Quality of life

The quality of life measured using the ULQIE resulted in higher values for the ASD and heart-healthy parents compared to the parents of a child with univentricular heart group ($p \leq 0.01$) for the overall scale, when all subscales are combined (Table 2).

"Satisfaction with the family" was comparatively similar in all three study groups ($p = 0.494$). All other subscales showed significant differences between parents of a child with univentricular heart and ASD parents. In terms of emotional stability and general well-being, the results differed within the subgroups of our study population. The univentricular heart parents were exposed to far greater emotional stress (emotional stability, MV = 2.42, SD = 0.929) compared to ASD parents (MV = 3.38, SD = 0.747), with a statistically significant mean difference of 0.96 (p-value < 0.001). However, two of the four items refer specifically to the illness of a child, so the results cannot be evaluated in comparison to the parents of heart-healthy children. "Physical functioning" was similar in the ASD (MV = 3.20, SD = 0.838) and heart-healthy (MV = 3.19, SD = 0.492) parents' group, but significantly lower for the parents of a child with univentricular heart (MV = 2.62, SD = 0.665, $p = 0.036$). Comparable results could be observed for the subscales of "well-being" and "self-development," where ASD parents and heart-healthy parents scored similarly and parents of a child with univentricular heart had significantly lower scores. See Figure 2 and Table 2 for a graphical representation of the subscales of the ULQIE questionnaire.

Needs

Comparing the needs for information on the disease and dealing with authorities of the univentricular heart (MV = 2.79) with the ASD parents' group (MV = 1.65), there is a statistically significant difference of 1.14 within the rating scale for needs (p-value < 0.001). This indicates that parents of a child with univentricular heart group had a significantly greater need for information about the disease, opportunities to talk, or support than parents of children with ASD.

Discussion

Psychosocial burden of disease

The impact of being diagnosed with univentricular heart on children has been studied several times, but the impact on the parents of the affected children has hardly ever been analysed, even though caregivers seem to know about the implications.[2,9,15] Studies have investigated the impact of a prenatal or postnatal diagnosis of complex CHD on parents, but not the long-term care of univentricular heart children.[17,18] Our study showed that there are significant differences between parents with an univentricular heart child, parents of children with a simple heart defect and parents with a child without a heart defect.

The results of the FaBel questionnaire, which measures the psychosocial burden of disease, showed a higher psychosocial stress and burden in univentricular heart parents. The highest burden was shown in the area "personal burden/future worries" for the parents of a child with univentricular heart. The "daily social

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load," "financial charges" and the "stress siblings" were also significantly higher in the univentricular heart than the ASD parents. This confirms the generally higher burden in univentricular heart parents than in the ASD comparison group. Only "coping problems" did not significantly differ between univentricular heart and ASD parents. An explanation for this could be, that the univentricular heart parents suffer from an overall higher burden in many areas but have good coping strategies to deal with this.

Wiedebusch et al. investigated the psychosocial burden and strains on parents of children with haemophilia, juvenile idiopathic arthritis and diabetes mellitus type 1 using the FaBel questionnaire.[16] The results are in concordance with results from the parents of a child with univentricular heart group in our study, which reported a similar burden of disease. These findings have also been confirmed in other studies, such as the cross-sectional study by Ravens-Sieberer et al., in which 273 families with chronically ill and disabled children were asked about their psychosocial burden of illness using the FaBel.[10] Jaschinski et al. investigated parents of children who received open-heart surgery (n = 113) with the conclusion, that the psychosocial impact depends on the number of operations. An increasing number of surgeries correlates negatively with the total burden measured by the FaBel ($R = -0.390$, $p < 0.001$). Furthermore, Jaschinski stresses the importance of "specialized psychological support" and the involvement of the whole family.[15]

The common factor in all these studies is the chronic nature of the disease. When we compared our univentricular heart to ASD parents, the latter reported a significantly lower psychosocial burden. This may be because univentricular heart parents still face far more limitations such as frequent hospital visits, limited mobility and uncertainty about the course of the disease. In addition, the continuous deterioration of cardiac performance that usually accompanies univentricular heart children with increasing age is also likely to play a role. This aspect is also confirmed by the worries from the parents about their children's future, since there is no corrective procedure, only a palliative treatment is possible for univentricular hearts.

Partnership satisfaction

The partnership satisfaction resulted in medium values for all three study groups for the global happiness assessment of the PFB. Overall, parents scored between rather happy and happy. Univentricular heart parents were similarly happy compared to parents in the heart-healthy group, whilst ASD parents were a little bit happier. Therefore, it can be deduced that all three groups considered their partnership to be quite good. This could be explained through good coping strategies of the univentricular heart parents. The importance of partnership support in terms of higher quality of life is also supported in other studies with chronically ill children by Wiedebusch et al.[13,16] Another key aspect that was identified by Biber et al. concerning the parental relationship: the importance of the perception of the burden, which differs between mother and father.[19] Therefore, parents of chronically ill children, which include parents of a child with univentricular hearts, should always be made aware of the importance of partnership cohesion.

Quality of life

In social sciences, both the objective standard of living and the subjective evaluation of the individual are incorporated into the understanding of quality of life.[20] Parents of chronically ill children often suffer from a particular burden in terms of time and personal resources. In order to be able to evaluate the family situation and the parents' concern about the child's state of health and future, the ULQIE represents a quality-of-life inventory specifically for groups of parents of chronically ill children.[11]

The quality of life showed a large difference between the univentricular heart, compared to the ASD parents and the heart-healthy parents on the other hand. This indicates a generally low impairment of quality of life for all three groups; however, the difference between the parents of a child with univentricular heart and the other two groups is significant. It must be taken into account that the ULQUIE is designed for the quality of life of parents of chronically ill children and therefore the values of the heart-healthy group can only be evaluated to a limited extent.

The highest values for all three study groups were found in the subscale "satisfaction with the family situation". This seems to indicate that families with children with univentricular heart do not seem to be negatively affected. The lowest values were obtained in the area of "self-development". Here, the parents of a child with univentricular heart group showed worse values compared to the ASD and the heart-healthy parents. Thus, in terms of self-actualization, the parents of a child with univentricular heart group faced greater limitations than the ASD and heart-healthy group.

"Emotional stability" was significantly different between the parents of a child with univentricular heart and ASD. These results may be criticized as not meaningful, because two of the four items relate specifically to a child's illness and not all families in the ASD group seem to perceive their child as chronically ill. Looking at the parents of a child with univentricular heart group alone, the medium results in the subscale of "emotional stability" can be explained by good coping strategies of the parents.

Another significant difference was found in the subscale "well-being". This confirms a higher stress of the univentricular heart parents in many areas of life. Why the heart-healthy group performed worse than the ASD group is not clear. However, the small sample size of the ASD and heart-healthy group should also be taken into account, which could be an explanation for this.

Comparing the quality of life results obtained on the basis of the ULQIE with those of other parents of chronically ill children reveals significant differences in certain areas, but also clear similarities. For example, Wiedebusch examined the quality of life of 285 parents of children with haemophilia, type 1 diabetes mellitus, and juvenile idiopathic arthritis.[16] Parents of children with juvenile idiopathic arthritis showed the highest impairment, followed by type 1 diabetes mellitus and parents of children with haemophilia. This corresponds to a moderate impairment of quality of life. Wiedebusch also investigated the parents of children with chronic renal failure, who showed a comparable overall ULQIE.[13] This is in concordance with our univentricular heart parents who achieved a mean value of 2.57 (SD = 0.658). Family satisfaction was high in all our study groups, which is consistent with results published by Wiedebusch et al. An impairment in self-development cannot just be observed in the group of the univentricular heart parents in our study, but was also reported by Wiedebusch et al. This reflects the severely limited opportunities to realize one's own needs in families with chronically ill children.[16]

Interesting comparisons can be drawn to a study by Lawoko et al., where parents of 1092 children with CHDs were compared to 112 parents of children with other diseases and 293 parents of healthy children.[21] Results for the overall quality of life measured

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by the "Göteborg Quality of Life Scale" showed a significant difference between parents with healthy children and parents of children with either CHDs or other diseases. This is in accordance with our results that show a small, but significant difference in overall quality of life for the three study groups. Interestingly enough, the parents of healthy children show only slightly higher life satisfaction than the parents of children with diseases. This supports our hypothesis that parents of children with diseases possess good coping strategies to cope with additional burdens.

Needs scale of parents of chronically ill children

There are various needs that may play an important role in the life of the affected families, like information regarding the disease, support and networking opportunities. One additional fundamental need is the necessity of time to yourself, the partner, other family members and friends. The overall needs scale showed a significantly higher need in the univentricular heart, compared to the ASD parents. This can be broken down into the areas of the need to receive more information regarding dealing with authorities and insurance companies and also the parents' desire for more time with their spouse/partner and the need for more information about possible therapies and learning more about possible diagnostic methods.

This is consistent with the 2008 work of Wiedebusch et al., who found a similar overall need for parents of children with JIA, diabetes mellitus Type 1 or haemophilia.[16] This is also echoed in a qualitative literature review by Fisher, who found three central themes of needs in parents of chronically ill children: the need for normality, the need for information and the need for relationships.[22]

Since more than half of the univentricular heart parents have a high need for more information on therapy and disease patterns and how to deal with authorities, this should be given even more consideration in the care of these parents and families by the responsible physicians, nursing staff and psychosocial staff. Likewise, the univentricular heart parents wished for more time with other family members, siblings and friends. A psychosocial contact person in the treatment centre was also very important for the parents of a child with univentricular heart group. Our results are comparable to those of parents with children with JIA, type 1 diabetes mellitus and haemophilia, who demonstrated high needs for information, support and time. This is in contrast with results from parents of children with ASD in our study, who had significantly lower needs.

Limitations

With 73 couples out of 217, we had an acceptable response rate of 33.6%. A larger number of participants would have been desirable, but difficult to implement under the given circumstances. Explanations for the response rate can be seen in time limitations, as the questionnaire was quite extensive with nine pages. A digital version of the questionnaire might have also increased the response rate. As the questionnaire was posted anonymously, no reminders could be sent to parents that had not answered. On the other hand, motivational factors may have played a role, especially for the PFB, which includes very private questions that certainly seemed too intimate to many parents despite adequate pre-information. Selection bias in the sample can also not be ruled out completely, as participation was voluntary and stress or the severity of the disease might have impacted participation. Due to the anonymization of the data collection, it is not possible to investigate whether the study participants differ from the parents who did not participate in some areas, such as psychological stability and educational background. Furthermore, the representativeness of the sample must also be discussed. The participant groups of parents of ASD children, as well as the children of parents with healthy hearts, were small in this study and larger samples are needed for a better comparison. Since the study participants were recruited monocentrically, the majority of them came from Munich or the surrounding area. Based on the reported educational status, living situation, work environment etc., the results seem to represent a sample of average middle-class families and the study groups did not significantly differ in relation to the parents' age, sex, marital status and number of children in the household. This is insofar significant, as financial resources and social milieu play a role in coping with illness and investigating the impact in families of all socioeconomic backgrounds is important.[23] On the other hand, it is important to note that children with an ASD were significantly younger than univentricular heart children. Furthermore, the cross-sectional design provides only a "snapshot" and a longitudinal study would be beneficial to investigate how the psychosocial burden changes over time.

Conclusion

On the one hand, our study confirms a higher psychosocial burden, limitations in daily life and family interactions in parents of univentricular heart children. On the other hand, a lower quality of life of parents of a child with univentricular heart compared to parents of children with ASD or no heart defect was observed. These differences also exist in comparison to families of children with other chronic diseases (e.g. haemophilia, type 1 diabetes mellitus and JIA) in other studies. It can be assumed that the general condition may deteriorate when children with univentricular heart reach adolescence and adulthood. Thus, univentricular heart means a lifelong exceptional situation not only for the affected person but also for their parents. Because of this, not only the patients themselves are exposed to special challenges and stresses throughout their lives but also their parents. This must be addressed in the psychosocial management of these families. One attempt to apply these results is the CHIP-Family study, where parents and children with CHD received additional psychosocial care to improve the well-being of the families, which could also be beneficial for parents of children with univentricular heart.[24]

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