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ORIGINAL RESEARCH

Impact of Raising Children with Rare Diseases on Parental Quality of Life and Family Functioning

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Abstract

Purpose: This study investigated how a child's functional ability and family cohesion impact the quality of life and family functioning of parents raising children with rare diseases.

Methods: Forty parents of children with Barth Syndrome (BTHS) or congenital muscular dystrophy (CMD) and forty parents of age-matched unaffected children participated in this study. Both groups of parents completed questionnaires providing information on their child's functional ability, family cohesion, parental quality of life, and family functioning.

Results: Multivariable general linear model results showed that parents of children with BTHS or CMD reported significantly lower quality of life and family functioning than those of unaffected children (-13.79, 95% CI: [-23.82, -3.75], p = 0.0078). In addition, a child's functional ability was found to be a crucial factor affecting parental quality of life and family functioning after controlling for child and parent variables (p = 0.0115).

Conclusions: This study laid the groundwork for understanding parents' experiences raising children with rare diseases. The findings of this study provide evidence for the need to develop effective strategies to support positive family functioning and well-being for families of children with rare diseases.

Keywords

Barth syndrome, Congenital muscular dystrophy, Family functioning, Quality of life, Rare disease

Introduction

Rare diseases are defined as small patient populations that affect less than 200,000 people in the United States [1]. Rare childhood diseases often have chronic impairing symptoms that require special care from family members or caregivers [2]. Barth syndrome (BTHS) and congenital muscular dystrophy (CMD) are rare diseases, which cause muscle fatigue and weakness from infancy or early childhood [3,4]. BTHS is an X-linked disorder found primarily in boys [5]. In the United States, BTHS occurs approximately 1 in every 1,000,000 births [6] and is characterized by cardiomyopathy, neutropenia, muscle weakness, growth delays, and exercise intolerance [7-9]. Children with BTHS demonstrate lower functional ability and health-related quality of life than unaffected children [10]. CMD is a group of heterogeneous muscular dystrophies exhibiting progressive muscle weakness [11]. The prevalence of CMD is 1 in 100,000 births [12]. The genetic mutation of muscle protein causes CMD, characterized by hypotonia and joint contracture [11,13]. Since no cure or specific treatment is yet available for these children, their families may need ongoing support to cope with the daily challenges they face.

Rare childhood diseases influence the everyday life of affected individuals as well as entire families [14-16]. Parents raising children with rare diseases have trouble managing their daily lives with the care giving roles [17,18] and have little knowledge about the life course of disease for their children, which causes anxiety [19]. In addition, having a child unable to engage in physical activities independently could place additional time constraints on other family members, as well as potentially generating conflict and tension [20]. Storch and colleagues [10] reported that parents of children with BTHS have significantly higher stress and distress levels than parents of unaffected children. Their stress and distress were related to care giving. Smith-Hoban

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and colleagues [21] conducted a caregiver assessment survey and found activities of daily living and strength of children with CMD had the greatest impact on the care giving burden. Recognizing the child's severity and chronic health condition completely changes the life perspective as well as quality of life of the entire family since they require a lot of support and care for their children [22]. In BTHS and CMD, however, the impact of affected individuals' health conditions on their parents' everyday life is less understood as well as parental quality of life.

Quality of Life (QoL) is defined as "an individual's perception of their position in life in the context of the culture and value systems in which they live and in relation to their goals, expectations, standards, and concerns" [23]. The poor physical and emotional health of the caregiver negatively affects the child, the family, and the community as a whole since it reduces the work productivity of the caregiver and raises healthcare costs both for the caregiver and the child [24]. Therefore, investigating the impact of a child's disease on quality of life of affected families is crucial to better understanding their circumstances and improving prospective support systems. Ammann-Schnell and colleagues [17] examined quality of life and life perspective of parents raising children with rare severe neurological disorders and found that the illness of the child had a considerable negative impact on the quality of family lives.

To date, most previous studies in BTHS and CMD have investigated the natural history and clinical features of these children. Evidence is limited regarding the impact of raising children with BTHS or CMD on families. The provision of this evidence is important for understanding parents' challenges in their daily lives and providing services and support for their needs. The Double ABCX model, which describes recovery from a child's disease and the process of adaptation of the families [25], is widely used for studies examining factors affecting families with children with disabilities [26-28]. The factors of the Double ABCX model encompass variables related to a child's disease, family, and healthcare services. Based on the Double ABCX model, this study hypothesized a child's functional ability and family cohesion as key factors and investigated the impact of these factors on parental quality of life and family functioning of parents raising children with BTHS or CMD.

Methods

Participants

Parents who have children or youth between the ages of 5 and 19 with BTHS (n = 20) or CMD (n = 20) (total N = 40) participated in this study. Parents of age-matched unaffected children (N = 40) served as controls. Inclusion criteria consisted of 1) Parents of children with BTHS, CMD, or unaffected children between the ages of 5 and 19 and 2) Parents who are English-speaking and able to read at an 8th-grade level. Parents of children with BTHS were recruited during their participation in the 7th International Scientific, Medical & Family Conference held by Barth Syndrome Foundation in Clearwater, FL. Parents of children with CMD were recruited by Cure CMD and CMD studies at the University of Florida (UF) and the National Institutes of Health. Parents of unaffected children were recruited by word of mouth and by flyers posted on UF department websites and Listserv, Face book groups, and Health Street (http://healthstreet.program.ufl.edu/). Both groups of parents completed a series of questionnaires in the cross-sectional study. This study was approved by the University of Florida Institutional Review Board. Written informed consent was obtained from each parent.

Instruments

The PedsQL™ Family Impact module (PedsQL FI): The PedsQL FI is a 36-item questionnaire that assesses the impact of a child's health conditions on parentalquality of life (QoL) and family functioning [29]. This instrument includes 8 scales: 1) Physical Functioning (6 items), 2) Emotional Functioning (5 items), 3) Social Functioning (4 items), 4) Cognitive Functioning (5 items), 5) Communication (3 items), 6) Worry (5 items), 7) Daily Activities (3 items), and 8) Family Relationships (5 items). Each item is scored on a 5-point scale (never a problem = 100; almost never = 75; sometimes = 50; often = 25; almost always = 0). The total score of the PedsQL FI is calculated using the mean of the 8 scales. Higher scores demonstrate better parental QoL and family functioning.

The Modified Barthel Index (MBI): The MBI is a 10-item questionnaire that measures the child's performance in activities of daily living (i.e., child's functional ability) [30]. The MBI consists of 10 scales 1) Personal Hygiene, 2) Bathing Self, 3) Feeding, 4) Toileting, 5) Stair Climbing, 6) Dressing, 7) Bowel Control, 8) Bladder Control, 9) Ambulation/Wheelchair, and 10) Chair/Bed Transfers. The MBI is administered by a therapist or family member/friend who can respond to the items based on their observation of daily life. Each item is scored from unable to perform task to fully independent (0~5 for Personal Hygiene and Bathing Self; 0~10 for Feeding, Toilet Transfer, Dressing, Bladder Control, Bowel Control, and Stair Climbing; 0~15 for Chair/Bed Transfer and Ambulation/Wheelchair). The total score is obtained by the sum of 10 items and ranges from 0 to 100. Higher scores indicate better functional ability or independence in activity of daily living.

The Family Adaptability and Cohesion Evaluation Scale-IV (FACES-IV): The FACES-IV is a 42-item questionnaire that measures the dimensions of family cohesion and family flexibility [31]. It consists of 6 scales: 1) Balanced Cohesion, 2) Balanced Flexibility, 3) Disengaged, 4) Enmeshed, 5) Rigid, 6) Chaotic. Each

item is scored on a 5-point scale (strongly disagree = 1; generally disagree = 2; undecided = 3; generally agree = 4; strongly agree = 5). The raw scores of subscales are converted to percentile scores using a percentile conversion chart. The total Circumplex Ratio is computed using these 6 subscales of the Cohesion and Flexibility scale. The formulas are presented below. The higher the Total Circumplex Ratio over 1, the more balanced/healthy the family.

Cohesion Ratio = Balanced Cohesion / ((Disengaged + Enmeshment)/2)

Flexibility Ratio = Balanced Flexibility / ((Rigid + Chaotic)/2)

Total Circumplex Ratio = (Cohesion Ratio + Flexibility Ratio)/2)

The demographic information form: The demographic information form, generated by the investigator of this study, consists of: 1) The child's date of birth and age, 2) Parent's age, 3) Marital status, 4) Race, 5) Employment, 6) Level of education, and 7) Family income.

Data analysis

An independent samples t-test was used for continuous variables, and the Chi-square test of association was used for categorical variables to analyze child and parent demographic variables. Given the observational nature of the study data, the investigator controlled for observed child and parent confounders by estimating a propensity score model for the probability of the rare disease group and unaffected group. The probability of each group was estimated with a logistic regression analysis that included observed child and parent covariates as explanatory variables. The estimated propensity score for each participant was then included in a general linear model to model an outcome variable, parental QoL and family functioning. In order to consider the impact of the analysis on the way the propensity score was accounted for, the investigator conducted a thorough sensitivity analysis by estimating this study's modeling results with a variety of different ways to include the propensity score.

Results

Eighty parents participated in this study, including 40 parents of children with BTHS (n = 20) or CMD (n = 20). Forty parents of unaffected children were selected as matched controls for comparison purposes. Table 1 displays summary statistics for all study variables.

Table 1: Summary statistics for study variables.

	Unaffected (n = 40)	Rare Disease (n = 40)	Total (N = 80)	*p*
	Count (%)	Count (%)	Count (%)	
Child Age				0.4957
> 10 years	25 (62.5)	22 (55.0)	47 (58.8)	
≤ 10 years	15 (37.5)	18 (45.0)	33 (41.2)	
Child Sex				1
Male	28 (70.0)	28 (70.0)	56 (70.0)	
Female	12 (30.0)	12 (30.0)	24 (30.0)	
Parent Race				< 0.0001
Non-Hispanic white	20 (50.0)	37 (92.5)	57 (71.2)	
Other	20 (50.0)	3 (7.5)	23 (28.8)	
Parent Education				< 0.0001
Graduate or professional degree	32 (80.0)	14 (35.0)	46 (57.5)	
Less than G/P	8 (20.0)	26 (65.0)	34 (42.5)	
Marital Status				0.1763
Married	33 (82.5)	37 (92.5)	70 (87.5)	
Other	7 (17.5)	3 (7.5)	10 (12.5)	
Employment				0.0049
Full-time	32 (80.0)	20 (50.0)	52 (65.0)	
Less than full-time	8 (20.0)	20 (50.0)	28 (35.0)	
Family Income				0.1709
$80,000 or above	21 (52.5)	27 (67.5)	48 (60.0)	
Less than $80,0000	19 (47.5)	13 (32.5)	32 (40.0)	
Child Functional Ability				< 0.0001
Total dependence	0 (0)	7 (17.5)	7 (8.8)	

Severe dependence	0(0)	1 (2.5)	1 (1.2)	
Moderate dependence	0(0)	13(32.5)	13 (16.3)	
Slight dependence	3 (7.5)	5 (12.5)	8 (10.0)	
Total independence	37 (92.5)	14 (35.0)	51 (63.7)	
	Mean (SD)	**Mean (SD)**	**Mean (SD)**	
Parent Age	41.5(6.4)	42.6(6.1)	42.0(6.3)	0.4239
Family Cohesion	2.7(0.8)	3.2(1.1)	2.9(1.0)	0.0122

$p < 0.05$; SD: Standard Deviation

Table 2: Summary of multivariable linear regression with continuous propensity score model results.

Variables	**Parameter Estimates**	**95% Confidence Interval**		***p***
		Lower	**Upper**	
Parental QoL and family functioning				
Rare	-13.79	-23.82	-3.75	0.0078
Unaffected	Reference			
Race				
Other	13	-26.22	52.22	0.5107
Non-Hispanic White	Reference			
Education				
Graduate/Professional Degree	2.59	-41.59	46.77	0.9072
Less than Graduate/Professional	Reference			
Income				
$80,000 or above	6.17	-6.11	18.51	0.3185
Less than $80,000	Reference			
Parent Age	0.06	-0.63	0.75	0.8605
Family Cohesion	3.97	-5.14	13.07	0.3879
Child Functional Ability				0.0115
Total dependence	-17.72	-31.64	-3.8	
Severe dependence	-34.4	-66.39	-2.41	
Moderate dependence	-18.08	-29.7	-6.45	
Slight dependence	-5.77	-18.72	7.18	
Independence	Reference			

$r^2 = 0.48$; $p < 0.05$

Results are stratified by the rare disease group versus the unaffected group. Statistical tests were applied to compare child and parent characteristics across the two groups. Parental race, education, and employment status, as well as child functional ability and family cohesion, were significantly different between the rare disease and unaffected groups. Parent race was substantially different ($p < 0.0001$), with 20 (50%) parents being non-Hispanic white in the unaffected group versus 37 (92.5%) in the rare disease group. Similarly, 32 (80%) of parents in the unaffected group had a graduate or professional degree, as compared to 14 (35%) in the rare disease group. The scores across the child's functional ability in the MBI indicated more dependency in the rare disease group ($p < 0.0001$). The family cohesion scores in the FACES-IV of the parents in the rare disease group were significantly higher than those in the unaffected group ($p = 0.0122$).

Table 2 shows modeling results for a multivariable general linear model. The outcome of parental QoL and family functioning was modeling as a function of child and parent characteristics. The propensity score was included in this model as a continuous covariate. The modeling results show that after controlling for child and parent factors, parental QoL and family functioning of the rare disease group was substantially lower than the unaffected group (-13.79, 95% CI: [-23.82, -3.75], $p = 0.0078$). In addition, the child's functional ability was a significant factor affecting parental QoL and family functioning ($p = 0.0115$). However, family cohesion was not a significant factor affecting parental QoL and family functioning after controlling for covariates.

Discussion

This study investigated the impact of a child's functional ability and family cohesion on parental

QoL and family functioning. The results indicated that parents in the rare disease group demonstrated significantly lower parental QoL and family functioning than those of the unaffected group, which is consistent with previous studies [17,24]. Parents in the rare disease group reported significantly higher tiredness during the day and difficulty finding time to finish household tasks than parents of unaffected children in this study. A child's functional ability, which indicates the severity of a child's disease, was a significant factor that affected parental QoL and family functioning in the parents of children with BTHS or CMD. The finding is consistent with a previous study that the severity of a child's function was a significant predictor of parental quality of life [32]. This is not surprising since BTHS and CMD are debilitating diseases that cause muscle weakness affecting overall physical functioning [10,11].

Parents in the rare disease group scored a higher mean of family cohesion than that of the unaffected group. However, after controlling for child and parent factors, family cohesion was not significantly associated with parental QoL and family functioning. Olson [31] mentioned that most scores of the FACES-IV range from 0 to 2. In this study, however, most parents from both groups scored higher than two on the FACES-IV. Since the parents maintained a high level of family cohesion, the data had less variability for comparing the impact between the two groups. The parents in this study had high levels of education and middle to upper-middle-class incomes, which might contribute to their heightened capacity to deal with the child's diseases [33]. Reichman and colleagues [20] mentioned that raising children with disabilities could positively affect the parents' inner strength and enhance family cohesion. By understanding their child's disability, parents could experience positive personality changes and improve relationships [34]. Therefore, the finding of this study may imply that parents of children with rare diseases become adjusted in managing their child's disease conditions.

For families who provide life-long care for their children, their burden of care may increase as the disease progresses [35]. Therefore, families require ongoing disease management support, up-to-date information, and education. In addition, they could benefit from counseling to enhance their adaptive skills to manage the increased burden and responsibilities [17,36]. Moreover, health professionals should consider providing family education and training, such as care giving techniques and strategies, encouraging families to become involved with activities outside the home, and connecting them with other families of children with disabilities for additional support [3,10,37]. Above all, to provide the most effective services for families of children with rare diseases, health professionals should understand the symptoms of rare diseases and the needs and experiences of the family so that affected individuals and families can receive efficient support and necessary information in a timely manner.

Limitations of this study include that many of the participants were recruited from rare disease associations, such as Barth Syndrome Foundation and Cure CMD, so findings cannot be generalized to other populations of children with rare diseases. Since these rare disease associations provide the important support (e.g., childcare information) and advocate for parents' needs, parents who joined associations might be better adapted to their child's condition and may maintain more balanced daily lives than those who do not. Additionally, most parents in this study were non-Hispanic White or Asian with middle to upper-middle-class incomes and high levels of education. Therefore, future research needs to include a more diverse sample of parents and children across a broader range of income, education, socioeconomic status, and racial groups.

Conclusion

By investigating the factors that affect the parents of children with rare diseases, this study sought to increase knowledge regarding how the child's functional ability and family cohesion relate to parental QoL and family functioning. The findings of this study contribute to laying the groundwork for understanding the experiences of parents raising their children with rare diseases. Health professionals should consider both the child and the family in providing holistic healthcare services and should strive to provide practical strategies to support positive family functioning and well-being in daily life.

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Disclosure Statement

The author reports no conflicts of interest.

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