Psychosocial Needs and Facilitators of Mothers Caring for Children with Duchenne/Becker Muscular Dystrophy

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Abstract:

Care guidelines for Duchenne/Becker muscular dystrophy (DBMD) include recommendations for professional assessment of caregivers followed by proactive psychosocial interventions. To inform neuromuscular care, this study described appraisals of psychosocial needs and caregiving facilitators of mothers of individuals with DMDB. Data were collected from U.S. mothers of individuals of DBMD using an online survey.

Two hundred and five mothers completed the survey. More than 50% endorsed unmet needs for managing uncertainty about the future and managing DBMD fears. Higher levels of unmet psychosocial needs were correlated with less disease progression in the child (rho= -0.166 p=0.02). Only 21% regularly used respite care and 57% worried about allowing others to care for their child. Highly-endorsed care facilitators included partner relationships (62.7%), child’s approach to life (58.7%), and family relationships (49.3%).

Our findings highlight the importance of social support to caregivers of children with DBMD. Starting when affected children are young, clinicians should assess caregivers’ unmet psychological needs, particularly their ability to cope with uncertainty and fear. Exploring needs and care facilitators using simple, focused questions may allow clinics to target and customize interventions. Interventions should help caregivers identify and build upon existing supports, and facilitate use of and comfort with respite care.

Keywords

Duchenne muscular dystrophy; Becker muscular dystrophy; caregiver; mothers; psychosocial needs; care facilitators; coping
1. Introduction

Children living with chronic illness have been shown to benefit when their care models address the psychosocial needs of the entire family [1]. This may be especially relevant for disorders such as Duchenne and Becker muscular dystrophy, where caregiving and financial demands increase over time and can impact the entire family [2-5]. Duchenne muscular dystrophy (DMD) is a rare, life-threatening disorder where affected individuals have progressive loss of functional muscle fibers that results in weakness, loss of ambulation that typically occurs in the teen years, and premature death [6,7]. Milder allelic forms of the disease include “intermediate” muscular dystrophy and Becker muscular dystrophy (BMD), which typically cause loss of ambulation at 13–16 years or over 16 years, respectively [6,7].

Care guidelines for DMD recommend family support that includes professional assessment of caregivers and families followed by proactive psychosocial interventions to meet their needs [6]. Though no specific methods or tools for caregiver and family assessment are recommended, a targeted evaluation of caregivers’ psychological and support needs may be practical and informative. As described by Boneskvi and colleagues [8], there are a number of advantages of needs assessment over other assessment methods: it 1) enables direct measure of individuals’ needs for help, which allows more direct indication of their resource needs; 2) allows the identification of the magnitude of needs, which makes it possible to prioritize interventions; and 3) enables the identification of individuals and subgroups with higher-level needs to facilitate early intervention.

The literature on parents caring for children with chronic disorders describes important components to include in a needs assessment. These include psychological responses to caregiving demands [9-13]; quality of family relationships and social supports [9, 14]; time for personal care and leisure activities [15,16]; impact of caregiving on ability to work a desired number of hours in a desired role [15-18]; and financial problems [14,18-20]. Similar to parents caring for children with other chronic
disorders, DBMD caregivers report higher than average rates of adverse psychological outcomes [3, 14, 21, 22] and lower quality of life [14]. Psychological outcomes have been found to be associated with factors such as child’s illness progression, social support, and financial burden [3, 4, 14, 21, 22]. Parents of children with DMD have been reported to be more likely than parents of healthy children to describe negative feelings about their child and their child’s outcomes [22]. Several other studies, however, have described components of DBMD caregiving that were perceived by caregivers to be important and rewarding [2, 23]. These studies also identify positive and proactive strategies to cope with the natural course of the disease and manage chronic sorrow [24]. This is consistent with data from other chronic disorder studies; while mothers were at increased risk for adjustment problems, the majority demonstrated good adjustment over time and reported adaptive coping [12].

1.1 Study Objective and Aims

The overall objective of the study is to identify potential targets for interventions to improve the psychological well-being of mothers caring for individuals with DBMD. This article describes the self-reported appraisals of caregiving facilitators and unmet psychosocial and respite needs of mothers of individuals with DMBD. Exploring these appraisals allows us to better understand the lived experience of caring for children with DBMD. The study aimed to provide a descriptive analysis of: a) psychosocial coping needs, b) respite care needs, and c) care facilitators. We also explored correlation between mothers’ psychosocial and respite needs and child’s ambulation status (used as a measure of stage of illness). It was hypothesized that later illness stages would be correlated with more unmet needs.

2. Materials and Methods
The survey items described here were purposively designed based on the existing literature and engagement with a DBMD community-based research team that comprises an expert parent advocate, DBMD clinicians, a psychotherapist, and social scientists. Items were refined through piloting with volunteers from the target population. Because the items were purposively developed, we included two open-ended items about needs and facilitators as an opportunistic way to assess face validity of listed items and whether any important domains were omitted. The questionnaire was implemented online using SurveyMonkey software.

This study was determined to be exempt by the Cincinnati Children's Hospital Medical Center Institutional Review Board.

Sociodemographic and Disease Characteristics: Participants’ age, ethnicity, education, marital status, income, and employment status were assessed. Additional items assessed demographic and disease characteristics of the child(ren) with DBMD, including age, diagnosis (Duchenne, Becker, or intermediary phenotype), and functional status, which was measured using a 7-item categorization largely focused on ability to ambulate.

Unmet Psychosocial Needs: This measure included eight items assessing a range of unmet psychosocial needs (see Table 1). The response option format was modelled on a similar unmet needs measure utilised in a previous study [25]. The items were summed and averaged for analyses (range 0-3). The unmet support measure had high internal consistency in this study with a Cronbach’s alpha coefficient of .91.

Respite Care Needs: Respite care items are showing in Table 2. Each item had 6-point Likert-type response options ranging from “strongly disagree” to “strongly agree”, as well as a “My child is independent” not applicable response option. Half of the items were positively oriented and half
negatively worded. The negatively worded questions were re-coded, N/A responses were removed, and items were summed. The Cronbach’s alpha coefficient was 0.73.

**Open-Ended Needs Item:** The survey included an open-ended question inquiring what participants perceived was the single most difficult aspect of caring for a child with DBMD.

**Caregiving Facilitators:** Six items, shown in Table 3, assessed factors that potentially facilitated mothers’ ability to provide “best care” to their child(ren) with DBMD. The 4-point response options ranged from “not at all” (1), to “very much” (4), as well as a “I do not have this type of relationship” response option.

**Open-Ended Self-Care Item:** The survey included an open-ended item asking about specific things participants do to help keep up their own wellbeing.

### 2.1 Participants and Recruitment

Participants were biological mothers who had at least one living child with Duchenne or Becker muscular dystrophy. Mothers lived in the United States, were 18 years of age or older, and able to answer questionnaires in English. Surveys were completed between November 2011 and October 2012.

Recruitment was conducted through online and e-mailed advertisements and social media postings through the Duchenne Connect Registry (www.duchenneconnect.org) and Parent Project Muscular Dystrophy, and continued through snowball recruitment using social media. Recruitment was also done using face-to-face invitations and advertisements distributed at Cincinnati Children’s Hospital Medical Center neuromuscular clinic. The total number of participants invited to the study is unknown given the variety of recruitment approaches, the use of social media, and overlap among populations in which recruitment occurred.
2.2 Statistical Analysis

Descriptive statistics were used to characterize the study sample based on demographics, unmet psychosocial, financial, and respite needs, and care facilitators. For open-ended questions, themes were identified and coded, and counts performed of the number of times each theme was mentioned. Spearman correlations were used to assess bivariate associations of baseline child's functional status with the summed scores of the psychological and respite needs measures.

3. Results

3.1 Sample

The analyses reported here were undertaken as part of a larger study, which also assessed the predictors of mothers’ psychological adjustment, the results of which has been reported separately [27]. Two hundred and five mothers completed the online survey. The majority (93.2%) identified as Caucasian. The mean age of the mothers was 44.0 years (SD=8.7), with a range of 27 to 71. The majority of participants had at least a college degree (136, 67.4%) and were employed or in a schooling program (145, 71.5%). The median household income was $50,000-$99,999. The majority of participants had one affected child (184, 89.8%). One hundred and seventy-four (84.9%) of the affected children had DMD, 23 (11.2%) had BMD, and 8 (3.9%) had an intermediate phenotype. The mean age of the affected child was 13.8 years (SD=7.2), with a range of 1 year to 40 years. Additional details about the population have been previously described [27].

3.2 Needs and Burden

Unmet Psychosocial Needs
Table 1 shows the proportion of mothers reporting unmet psychosocial support needs. The three most frequently endorsed needs, with more than 50% responding medium or high need, were: specific ways to deal with uncertainty about the future, manage fears related to DBMD, and cope with being a mother of a child with DBMD. The mean averaged score on the unmet psychosocial needs measure was $M=1.3$ (SD=0.7, range 0 to 3). The child’s functional status was significantly and negatively correlated with the average needs score (Spearman’s rho= -0.166, p= 0.02).

**Respite Needs**

Across all of the respite items (see Table 2), approximately 30% of the participants rated their child as independent, making the question not applicable. Of the remaining participants for whom the questions were applicable, 26 (21%) agreed or strongly agreed to regularly using respite care, and 37 (27%) agreed or strongly agreed with having all of the information they need to find respite care. Seventy-six (57%) agreed or strongly agreed that they worried about allowing others to care for their child, and 70 (53%) agreed or strongly agreed that their child was willing to be cared for by someone else. Ninety four (68%) agreed or strongly agreed that they could benefit from a break, while 21 (17%) agreed or strongly agreed that finding respite care was more trouble than it is worth. The child’s functional status was not significantly correlated with the summed respite measure (Spearman’s rho= -0.87, p= 0.14).

**Open-ended needs question**

One hundred and twenty-one participants provided an open-ended response to the question, “Right now, is there one particular thing about DBMD that makes it most difficult for you to care for your child?” The four most common themes comprised unmet psychosocial needs (n= 59), access barriers (n= 46), physical demands of caregiving (22), and negative effects of the child’s behavioral or psychological issues (20). Mothers described unmet psychosocial needs regarding managing
uncertainty about future decline, bearing witness to disease progression, and helping their son deal with his progressing illness. Access issues that mothers described included access to quality care, equipment, and respite services.

3.3 **Personal Strength Appraisals**

*Care Facilitators*

Participants reported many factors that facilitated provision of the “best care” for their child (see Table 3). Greater than 50% of participants responded “somewhat” or “very much” to each of the care facilitators provided. The items with the highest endorsement of “very much” were partner relationships (62.7%, n=126), the child’s approach to life (58.7%, n=118), family relationships (49.3%, n=99) and the mothers’ faith/spiritual beliefs (40.8%, n=82).

*Open-ended self-care question*

Participants were asked, “What specific things do you do, if any, to help keep up your own wellbeing?” Of the 172 open-ended responses provided, the majority described social support (n=107) and exercising (n=75). Another group of frequently described self-care activities (n=62) included “taking time out” for hobbies, time alone, writing/bloggings, gardening, reading books, etc.; as described by several mothers, doing “normal things.” Twenty-two women responded that they were unsuccessful or did nothing to keep up their own wellbeing.

4.0 **Discussion**

This study provides a description of the psychosocial support needs and caregiving facilitators of mothers of individuals with DBMD. The results inform clinical care provision and may represent an opportunity to extend the role of the genetic counselor through neuromuscular specialty clinics. The least-met psychosocial needs were coping with challenging emotions: dealing with uncertainty about
the future, and fears related to DBMD. Open-ended responses reinforce the importance of managing negative feelings. Contrary to our hypothesis, higher psychosocial support needs were associated with earlier (less severe) disease stages. This suggests that over time, mothers perceive that more of their needs are met and feel better equipped to cope with DBMD using their internal and external resources.

Participants identified many areas of caregiving strength. Support from a partner and family members was important to facilitate the best care, as was the child’s approach to life. More than half of respondents also endorsed faith/spiritual beliefs and support from healthcare providers as care facilitators. Open-ended comments highlight exercise, social support, and taking “time out” for pleasurable activities as important components to self-care. These facilitators provide important targets for interventions to improve mothers’ care-related self efficacy.

Our strong, recurring data on the importance of social support to caregivers of children with DBMD is consistent with other studies [2,3]. Access to a full complement of social support, however, may presuppose the caregiver’s ability to “get away” from caregiving responsibilities. A review by Robertson and colleagues [28] reports benefits of respite that include improving caregiver, sibling, and affected child wellbeing. Our participants, however, reported relatively infrequent use of respite care, despite a desire for respite and a perceived willingness of the affected child to accept such care. Our respite data are complicated by almost a third of participants indicating that the questions were not applicable because their child is independent, which is unexpected. This may reflect variability in knowledge about and access to respite care, or an unclear question set. The wide age range of the affected children for whom mothers choose N/A suggests a need for additional research to understand mothers’ experiences with and perceptions of respite.
The open-ended questions provide potential topics for downstream research that include care and service barriers, the physical demands of caregiving, and specific needs of caregivers whose children with DBMD have serious behavioral or psychological issues. Though preceding survey questions may have influenced participants' thinking toward psychosocial needs and social supports, it is striking how few mothers described information seeking as a coping mechanism in the open-ended questions. While seeking information about care and management is undoubtedly a coping mechanism used by parents of children with DBMD [24], healthcare providers are likely to be more adept at addressing such information-focused needs and many information sources are available to parents, as compared with less easily addressed emotion-focused needs.

Limitations

The mothers who participated in this study were recruited, in part, from DBMD community resources that members opt to join. It is possible that unmet needs and care facilitators are different in scope and relative importance in this group than among other mothers managing children with DBMD. The study is limited by the study sample’s high rate of married participants, as well as the relatively high employment level and income that is above the U.S. median household income [29]. This may impact the generalizability and interpretation of some findings. Finally, the response rate cannot be evaluated given the variety of recruitment approaches, the use of social media and snowball recruiting, and overlap among the recruitment populations.

Clinical Implications
Clinical teams in neuromuscular centers should aim to systematically explore and address psychological needs of all caregivers, starting when affected children are very young, including coping with DBMD-related uncertainty and fear. With regard to respite care, clinicians should anticipate that mothers may worry about allowing their children to be cared for by someone else, and support mothers in achieving higher levels of comfort. Among the neuromuscular clinical care team, genetic counselors may be particularly suited to engage in these longitudinal assessments and explorations of psychosocial needs.

Exploring caregivers’ needs and care facilitators using simple targeted questions, similar to the questions used in this study, may allow clinic teams to customize interventions based on specific needs. These discussions might best occur away from the affected child to allow mothers to explore fully explore their feelings and describe their needs without concern for upsetting the child. Psychosocial interventions may help caregivers identify and building upon existing supports and facilitators, which may lead to better psychosocial outcomes in the patient and entire family that is managing the effects of DBMD.

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References


