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Mothers' unmet support needs and psychological adaptation to Duchenne/ Becker muscular dystrophy

CHAPTER 2.

Mothers' unmet support needs and psychological adaptation to Duchenne/Becker muscular dystrophy

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ABSTRACT

Duchenne and Becker muscular dystrophy (DBMD) cause significant emotional and care-related burden on caregivers, but studies have not assessed predictors of disease-specific adaptation. Using a community-engaged approach focused on supporting mothers in positive aspects of caregiving, this study aims to assess mothers' unmet needs and identify predictors of mothers' psychological adaptation to DBMD.

Mothers of at least one living child with DBMD completed an online survey (n=205) and a two-year follow up (n=144). The surveys measured unmet needs, DBMD-related adaptation, optimism, resilience, perceptions of caregiving, and child's functional status.

The greatest unmet support needs were in managing future uncertainty and DBMD fears. Unmet needs were modestly but significantly higher for mothers of ambulatory children (M=1.43) than nonambulatory (M=1.12), $p=0.02$. Mothers indicated a desire for more information on respite care (40%). Increased psychological adaptation was predicted by resilience ($\beta=.264$, $p=.001$) and perceived positive impact ($\beta=.310$, $p<.001$), controlling for mother's age ($\beta=-.305$, $p<.001$). Child's functional status did not predict adaptation.

Clinicians should address unmet support and respite needs. Though increased caregiver burden is anticipated with disorder progression, burden did not predict psychological adaptation. Efforts to improve wellbeing should instead focus on fostering resilience and benefit finding, especially as mothers age. Additional exploration is needed to better understand the effects of increasing mother's age and worsening child's functional status on support needs and psychological adaptation.

INTRODUCTION

Duchenne and Becker muscular dystrophy (DBMD) are rare, progressive, X-linked diseases of muscle wasting.¹ Duchenne muscular dystrophy is the more common and severe of the two disorders, with noticeable symptoms in early pediatric years.² It leads to severe progressive muscle weakness that results in increasing care needs as the child ages, and death typically in the late 20s.³ Becker muscular dystrophy is more heterogeneous, ranging from a course similar to Duchenne to milder presentations with later onset and more slowly progressing weakness.²

Several cross-sectional studies have explored the impact of caring for a child with DBMD on parent/guardian caregivers. Studies have shown high caregiving demands and high perceived burden,⁴⁻⁷ stress,^{5,7,8} distress,^{5,9} and lower health-related quality of life (QoL) in caregivers.¹⁰

Psychological outcomes have been found to be associated with factors such as child's illness progression, social support, and financial burden.^{4,5,9,10}

However, survey and interview studies have also described components of DBMD caregiving that were perceived as beneficial, including an improved ability to appreciate life experiences and other positive coping strategies to manage the progressive disease course and chronic sorrow.¹¹⁻¹⁴ Pangalila and colleagues' (2012) cross-sectional study of parents of adults with DBMD found high subjective burden, but also high rating of care as important and rewarding.⁴ Kenneson and Bobo⁵ found that general life satisfaction was associated with high social support, high resiliency, and high income. While Hatzmann and colleagues assessed factors associated with health-related quality of life,¹⁰ QoL is not specific to the caregiving experience and includes both positive and negative domains. Further, causation cannot be assessed from the existing cross-sectional studies. No longitudinal studies have been reported that are able to identify predictors of disease-specific caregiver wellbeing.

Studies of caregivers of other chronic disorder populations provide additional insight into wellbeing. While at increased risk for adjustment problems, the majority of caregivers demonstrated good adjustment and reported adaptive coping.¹⁶ The importance of positive psychological responses to caregiving demands;¹⁷⁻²⁰ quality of family relationships and social supports;^{10,18} time outside the home;²¹⁻²⁴ and financial resources^{10, 24} have been demonstrated.

Though mothers caring for children with DBMD experience challenges to their wellbeing, unmet caregiving needs and DBMD adaptation can be explored in a positive framework of coping, optimism, and resilience. Disorder-specific adaptation is a wellbeing concept defined as a dynamic process of coming to terms with the implications of a health threat and the outcomes of that process.²⁵ Our choice of psychological adaptation to DBMD as a study outcome measure reflects the positive personal impact reported by caregivers and is consistent with the preferences expressed during community engagement (described below). Predictor variables were chosen based on the literature and through community engagement, and are described further under the "Measures" section.

Aims and Hypotheses

This study aims to: (i) assess mothers' unmet support, respite, and financial needs; (ii) determine the impact of the child's functional status on mothers' unmet support needs; and (ii) to determine

whether psychosocial adaptation to DBM two years later can be predicted by baseline child's functional status, mothers' dispositional optimism, resilience, perceptions of DBMD caregiving, and their coping self-efficacy.

It was hypothesized the child's functioning at the time of the baseline survey would be associated with mothers' unmet support needs and psychological adaptation—specifically, that worse child function (i.e., more severe illness) would be associated with more unmet needs and negative impacts on adaptation. It was also hypothesized that mothers' psychological adaptation as measured two years after the baseline survey would be associated with higher levels of dispositional optimism, resilience, perceived positive impact of DBMD, perceived control, and coping efficacy, and with lower levels of worry about care and perceived caregiver burden.

Longitudinal data collection is advantageous because it facilitates understanding of causality in assessing predictors of psychological adaptation. Such data will inform the development of interventions aimed at improving caregivers' wellbeing, by identifying when interventions may be most necessary and which caregivers are most likely to develop adverse psychological outcomes so that interventions can be targeted more precisely.

MATERIALS AND METHODS

Approach

This study used a community-engaged research approach, in which advocates, clinicians, caregivers, and social science researchers identified the research agenda, design and delivery.^{26,27} Most notably, the focus of the project is responsive to a community-identified need to engage in needs assessment while appreciating and exploring positive perceptions and outcomes experienced by caregivers. The long-term objective is to develop interventions focused on meeting needs and improving adaptation rather than solely target negative impact on caregivers.

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

Participants

Participants were biological mothers of at least one living child with Duchenne or Becker

muscular dystrophy, who lived in the United States, were 18 years of age or older, and able to answer questionnaires in English. The online questionnaire was implemented using SurveyMonkey software. Participants completed the baseline survey between November 2011 and October 2012, followed by a follow up survey that was distributed two years later.

Recruitment strategy

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; and through 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 impossible to calculate given the variety of recruitment approaches, the use of social media and snowball recruiting, and overlap among the recruitment populations.

Procedure

Eligible mothers provided their contact information and each participant was asked to complete a baseline questionnaire. With the exception of two participants who requested mailed paper copies, the participants responded to surveys online using unique survey links for each participant. The unique link to a follow-up survey was sent by email at about 24 months after the baseline survey.

Measures

Demographic/Disease Characteristics: Participants' age, ethnicity, education, marital status, income, employment status, state of residence and mother's carrier status were assessed. Additional items related to the child included: diagnosis (Duchenne, Becker, or intermediary phenotype), age, age at diagnosis, and functional status. The latter was measured using a 7-item categorization where a higher score means worse condition. The same functional assessment item has been used in the DuchenneConnect patient registry and is an adaptation of the stages in the Duchenne care guidelines.¹

Personal Attributes

Dispositional optimism was measured with the 10-item Life Orientation Test Revised (LOT-R).²⁸ Dispositional optimism is associated with psychological wellbeing and physical health across a range of disease populations.²⁹ Cronbach's alpha in this sample was 0.87. **Resilience**

was measured using the Resilience Scale for Adults (RSA), which measures 'protective resources' that have been demonstrated to facilitate flexibility in coping.³⁰ Cronbach's alpha was 0.94.

Perceptions of DBMD Caregiving

Perceived Caregiver Burden was measured using the 12-item Zarit Burden Interview (ZBI).³¹ A score of 17 or above may be used as cut off point to identify high burden.³¹ In this sample, Cronbach's alpha was 0.89. **Perceived Personal Control** was measured using five questions about control over DBMD in general, daily symptoms, long-term course, medical care and treatment, and control by others (adapted from Lipinski and colleagues).³² Cronbach's alpha was 0.79. **Worry about Care for Child with DBMD** was measured using three items purposively designed to assess amount, frequency, and intensity of DBMD-specific care worry (developed as suggested by McCaul and Goetz).³³ Cronbach's alpha was 0.89. **Perceived Positive Impact** was measured with one item purposively developed for the study ("How much of a positive effect does your child's condition have on your entire family?"). **Coping Self-Efficacy** was assessed with the **Coping Self-Efficacy Scale (CSES)**, a 26-item measure of perceived self-efficacy for coping with challenges and threats.³⁴ Cronbach's alpha was 0.97.

Mothers' Unmet Needs

The unmet needs items were purposively designed based on the literature and through community engagement.

Unmet Support Needs includes eight items that assess a range of unmet needs related to coping with DBDM (see Table 2). It is modelled on a needs measure utilised in a previous study.³⁵ It had high internal consistency with a Cronbach's alpha coefficient of .91. **Financial Need** was measured with one item, "If I had more money I could better care for my child with DBMD" scored on a 4 point Likert-type scale. The **Respite Care Needs** items range from frequency of use, information needs, attitudes, worry, and child willingness (Table 3). Each item had 6-point Likert-type responses and a "My child is independent" option. The item "I could benefit from a break from caring for my child" reduced the internal consistency and was removed from the summed score. The Cronbach's alpha was 0.73.

Psychological Adaptation to DBMD

The outcome variable, mothers' **psychological adaptation** to DBMD, was measured with the 20-item psychological adaptation scale (PAS), which is designed to measure adaptation to a

chronic condition or disease risk by patients or caregivers.³⁶ Cronbach's alpha was 0.96.

Statistical analyses

Data were initially explored with descriptive statistics and graphs. Separate one-way, between-groups analyses of variances (ANOVA) with approximation for homogeneity of variance, when appropriate, were conducted to assess whether child's functional status (ambulatory, children in transition to the use of a wheelchair full-time, or non-ambulatory) was associated with differences in unmet needs. For the ANOVA analyses only, to allow sufficient sample sizes the baseline child function item was re-coded into three ambulation categories: ambulatory children, children in transition to the use of a power wheelchair full-time, and full-time users of power wheelchairs.

We examined bivariate relationships among variables measured at baseline (child's functional status, dispositional optimism, worry about care, perceived control, caregiver burden, resilience, coping efficacy, and unmet support needs) and the outcome variable measured at the two-year follow up (mothers' psychological adaptation to DBMD). To assess predictors of psychological adaptation, all predictor variables with $p < 0.25$ in the bivariate analysis were entered into a multiple linear regression, then progressively eliminated until only those with p-values of < 0.05 remained. Potential confounders (mother's age, income, carrier status) were then entered one at a time and retained in the regression if the β associated with any of the predictor variables changed by more than 10%.

Prior to analysis, child's functional status was chosen to include in analyses rather than child's age or diagnosis. The clinical variability in the DBMD diagnostic categories^{1,2} makes anticipation of natural history or stage based on age difficult. Further, given the lower prevalence of Becker muscular dystrophy, it was likely there would be an insufficient sample size for analyses in that subgroup.

RESULTS

Sample

Two hundred and five mothers participated in the baseline survey, and 144 participated in the follow up survey two years later (a 30% loss rate from the baseline survey cohort). This includes

two mothers who completed the baseline survey but did not complete the 2-year follow up because their affected child died between survey points.

At the time of the baseline survey, one hundred and ninety-two (93.2%) identified as Caucasian, 11 (5.3%) as Hispanic, 6 (2.9%) as Asian, and 7 (3.4%) as “other”; respondents had the option of endorsing more than one category. 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 was employed or attending school part- or full-time (145, 71.5%). The median household income was \$50,000-\$99,999. One hundred and seventy-seven (86.3%) were married or in a long-term committed relationship, 24 (11.7%) were divorced or separated; 3 (1.5%) had never married, and 1 (0.5%) was widowed. Ninety-six participants (46.8%) reported being DBMD carriers; 78 (38.0%) were non- carriers; and 31 (15.1%) did not know whether they were carriers.

The majority of participants had one affected child (184, 89.8%), 19 (9.3%) had two affected children, and two (1.0%) had three affected children. One hundred and seventy-four (84.9%) of the affected children had DMD, 23 (11.2%) had BMD, and eight (3.9%) had an intermediate phenotype. The mean age of the affected child was 13.8 years ($SD=7.2$), with a range of one year to 40 years.

Mean child functional status was 3.5 ($SD= 1.8$, $N=205$) at baseline and 3.9 ($SD=1.8$, $N=144$) at 2-year follow up, with higher numbers indicating worse function. Using baseline data, the 7-item child functional categorization was re-coded into three ambulation categories: ambulatory children (83, 40.5%), children in transition to the use of a power wheelchair full-time (48, 24.4%), and full-time users of power wheelchairs (74, 36.1%). If the participant had more than one affected child, the functional status of the oldest living child is reported.

There were no statistically-significant differences in median income, mean age, or mean child functional status between those who answered the first survey only and those who answered both the baseline survey and survey at year 2.

Personal Attributes and Perceptions of DBMD Caregiving

Table 1 presents means and standard deviations of mothers' perceptions of BDMD caregiving and their personal attributes. Defining a high ZBI score as 17 or above,³¹ 48.2% of the mothers

reported high burden at baseline and 52.8% of the mothers reported high burden at two-year follow up.

Table 1 Measure/Item Means: Baseline and 2-Year Follow Up

	Scale/Item Range	Baseline Mean (n=205)	2 Year Mean (n=144)
Child function	1-7	3.5 (SD= 1.8)	3.9 (SD=1.8)
LOT-R	0-24	14.3 (SD= 4.6)	—
RSA	1-5	3.9 (SD= 0.5)	3.9 (SD=0.5)
Zarit burden	0-48	17.1 (SD= 8.6)	17.7 (SD=8.6)
Control	1-11	5.5 (SD= 2.1)	4.6 (SD=2.1)
Worry	3-15	7.3 (SD= 2.9)	—
Positive impact	0-10	5.6 (SD= 2.9)	—
Coping self efficacy	0-260	156.5 (SD= 51.6)	168.4 (SD= 49.4)
PAS	1-5	3.5 (SD=0.9)	3.6 (SD=1.0)
Household income	<\$50,000 to ≥\$250,000	Median \$50,000 - \$99,999	Median \$50,000 - \$99,999

Mothers' Needs

Unmet Support Needs

Table 2 shows the proportion of mothers reporting unmet support needs, as measured in the baseline survey. 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; specific ways to manage fears related to DBMD; and specific ways to cope with being a mother of a child with DBMD. The mean score on the unmet psychosocial needs measure was $M=1.31$ ($SD=0.7$, range 0 to 3). A modest but significant difference in mean needs scores was observed depending on the child's functional status, $F(2, 200)=4.0$, $p=0.02$. Post-hoc comparisons indicated that the mean score for the ambulatory children ($M=1.43$, $SD=0.6$) was significantly higher than the mean for the non-ambulatory children ($M=1.12$ $SD=0.8$). The transition to wheelchair group ($M=1.38$ $SD= 0.7$) did not differ significantly from either the ambulatory or the non-ambulatory group.

Table 2. Mothers' Unmet Psychosocial Needs

	Don't have this need	Use to have this need but not anymore	Low need	Medium need	High need
Specific ways to deal with uncertainty about my child's future	6.4% (13)	6.9% (14)	25.1% (51)	34.0% (69)	27.6% (56)
Specific ways to manage my fears related to my child's DBMD	7.4% (15)	9.4% (19)	28.1% (57)	34.0% (69)	21.2% (43)
Specific ways to cope with being a mother of a child with DBMD	6.9% (14)	17.7% (36)	22.2% (45)	32.0% (65)	21.2% (43)
Better ways to get the support I need from others	11.8% (24)	8.4% (17)	28.1% (57)	30.0% (61)	21.7% (44)
Ways of self-care that improve my sense of wellbeing and happiness	14.8% (30)	8.9% (18)	24.6% (50)	31.5% (64)	20.2% (41)
Specific ways to manage my sadness related to my child's DBMD	7.4% (15)	18.2% (37)	25.1% (51)	28.1% (57)	21.2% (43)
How to take time for healthy life choices (such as diet and exercise)	12.8% (30)	11.3% (23)	26.6% (54)	27.6% (56)	21.7% (44)
Whether the way I feel is normal	23.6% (48)	16.7% (34)	30.5% (62)	20.2% (41)	8.9% (18)

Financial Needs

In response to the item eliciting perceived financial burden, the median was 3.0 and mean was 2.76 (SD=1.17, range 1-4) and 60.7% (122) indicated “somewhat” or “very much”. No significant differences were observed in the perceived financial burden, $F(2, 198) = 2.8$, $p=0.06$, based on the child’s functional status.

Respite Needs

Across all of the respite items (see Table 3), 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%) 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%) that their child was willing to be cared for by someone else. On the summed score, no significant differences were observed in the respite needs, $F(2, 46) = .61$, $p=0.55$, based on the child’s functional status.

Table 3. Mothers' Respite Care Needs

	Strongly disagree	Disagree	Neutral	Agree	Strongly agree	N/A child is in-dependen t
I have all the information I need on how to find respite care	14.9% (30)	24.8% (50)	10.9% (22)	11.9% (24)	6.4% (13)	31.2% (63)
I regularly use respite care	23.8% (48)	16.3% (33)	7.4% (15)	8.4% (17)	4.5% (9)	39.6% (80)
Finding respite care is more trouble than it is worth*	8.9% (18)	13.4% (27)	30.2% (61)	5.9% (12)	4.5% (9)	37.1% (75)
I am worried about allowing someone else to care for my child*	5.4% (11)	14.9% (30)	8.4% (17)	22.3% (45)	15.3% (31)	33.7% (68)
My child is willing to be cared for by someone else	5.9% (12)	11.9% (24)	13.4% (27)	21.3% (43)	13.4% (27)	34.2% (69)
I could benefit from a break from caring for my child	2.5% (5)	4.5% (9)	14.9% (30)	24.3% (49)	22.3% (45)	31.7% (64)
I don't deserve a break from caring for my child*	34.2% (69)	19.8% (40)	9.9% (20)	3.0% (6)	1.5% (3)	31.7% (64)

* Items are reverse scored in calculation of summed score

Bivariate analyses

Bivariate analysis of predictor variables and unmet support needs (measured at baseline), and psychological adaptation (measured at two-year follow up), showed statistically-significant, positive relationships between the psychological adaptation and the predictors dispositional

optimism, resilience, perceived control, positive impact, and coping self efficacy. Higher mothers' unmet support needs were associated with lower dispositional optimism, resilience, perceived control, and coping self-efficacy; and higher perceived burden. Worse child's functional status was significantly associated with lower unmet support needs and resilience; and higher perceived caregiver burden and perceived positive impact (see Table 4).

Table 4 Pearson Correlations Among Needs, Predictors, and Psychological Adaptation (PAS)

Scale/Item	Unmet support needs	PAS (2 year follow up)	Child function	Mother age
Unmet support needs	—	-.092	-.162*	-.232*
PAS	-.092	—	.006	-.336**
Child function	-.162*	.006	—	.494**
Mother age	-.232*	-.336**	.494**	—
Household income [^]	-.167*	-.133	-.060	.233**
LOT-R	-.355**	.190 ⁺	.042	.197**
RSA	-.391**	.330**	-.166 ⁺	-.002
Zarit burden	.307**	-.117	.312**	.087
Control	-.210**	.229 ⁺	.042	-.097
Worry	.453**	-.084	.091	-.044
Positive impact	-.089	.399**	.170 ⁺	-.033
Coping self efficacy	-.502**	.348**	.009	.109

[^] Spearman rho

* Correlation is significant at the 0.05 level (2-tailed).

** Correlation is significant at the 0.01 level (2-tailed).

Multivariate analyses of psychological adaptation

A multiple linear regression was performed to assess predictors of psychological adaptation. The final model (see Table 5) included perceived positive impact (beta = .310, $p < .001$) and resilience (beta = .264, $p = .001$) and which explained 30.5% of the variance in mothers' psychological adaptation to DBMD, after controlling for effects of participants' age and income. Worry about child's care, perceived control, coping self-efficacy and perceived caregiver burden were no longer significantly associated with psychological adjustment.

Table 5: Predictors of Psychological Adaptation at 2 Year Follow-up (N=136)

<i>Variable</i>	<i>Beta coefficient</i>	<i>T</i>	<i>p</i>
Household income	-.088	-1.169	.245
Mothers' age	-.305	-1.169	$p < .001$
Perceived positive impact	.310	4.122	$p < .001$
Resilience	.264	3.459	$P = .001$

Final model: $R^2 = 0.325$ $F(4, 136) = 15.889$, $p < .001$. Adjusted $R^2 = 0.305$, $R = 0.570$

DISCUSSION

This study provides clinically-relevant data about mothers' needs, strengths, and adaptation to caregiving for a child with DBMD. The use of psychological adaptation to DBMD as our primary outcome reflected stakeholders' attitudes that caring for a child with DBMD comes with benefits, and clinical interventions should focus on addressing needs and fostering positive outcomes. Greater resilience and positive impact predicted better psychological adaptation in mothers. Resilience is a multidimensional personal attribute that may be shaped by personality traits, evolving appraisals, social support systems, and family environments, and it is likely to be responsive to interventions.³⁷⁻³⁹ Positive impact is a representation of benefit finding—a perception that major positive changes can come from a traumatic life experience.⁴⁰

Consistent with previously-reported studies^{5,6} we found that worse functional status in the child was associated with higher perceived caregiver burden, and approximately 50% of mothers reported a high level of burden. Higher perceived burden was also associated with higher unmet support needs. Although families, clinicians, and other stakeholders might anticipate increased burden to mothers with disorder progression, this study showed that burden did not predict

psychological adaptation, suggesting that mothers manage increasing caregiving burden over time. Interventions targeted at caregiver burden might not be the most effective methods to improve mothers' psychological wellbeing.

With regard to mothers' needs, the least-met support needs were coping with challenging emotions: dealing with uncertainty about the future, and fears related to DBMD. Many clinicians may benefit from further training to address these areas. In addition, we identified a need to improve knowledge about, and use of, respite care. While research underscores the importance of social support,^{4,5} access to a full complement of social support may presuppose the caregiver's ability to "get away" from caregiving responsibilities. Demonstrated benefits of respite include improving caregiver, sibling, and affected child wellbeing.⁴¹ However, our participants 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 respite and access 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.

We report unexpected relationships among mother's age, child's disease progression, unmet support needs, and adaptation. A higher score on unmet support needs was modestly but significantly associated with less disease progression in the child, and was correlated with younger mothers' ages. Higher psychological adaptation to DBMD was predicted by younger mothers' age. However, child's functional status was not a predictor of psychological adaptation. While mothers may be able to attribute more benefit to the DBMD experience and perceive that more of their support needs are met as their children's symptoms become more advanced, their resilience may be challenged as they age. It is also possible that there is a response shift in the function measure, where mothers of children in earlier stages provide more optimistic reports of their child's functional status than mothers of children progressing to later stages of DBMD. Additional exploration is needed to better understand the effects of increasing mother's age and worsening child's functional status on support needs and psychological adaptation.

Though the study sample was broadly recruited through diverse sources, possibly increasing the representativeness of the sample, the study is limited by the opt-in nature that may have

generated participation bias. This could not be evaluated, as details on individuals who did not participate were not available. In addition, 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.

Though there was participant loss between the baseline and the survey two years later, there were no significant differences in income, age, and child functional status among those who participated only at baseline and those who participated two years later.

Traditionally underserved minority and low SES populations were under-represented in the survey, as were non-married mothers, which may have implications for the generalizability of the findings. For example, while the data do not support an increase in financial concerns as the disease progressed, most participants indicated that they would be able to better care for their children with more money. A more representative sample may have greater financial concern. Financial concerns may become more significant in the future, once new therapeutics (many of which are anticipated to be associated with high annual cost, similar to drugs for other rare disorders)⁴² are approved. The study should be repeated in a generalizable clinic-based sample to strengthen the basis for clinical recommendations.

Clinical Implications

Care guidelines for DMD recommend family support that includes professional assessment of caregivers and families followed by proactive psychosocial interventions to meet their needs.¹ Our results highlight the need for systematic exploration of caregivers' unmet support needs, especially those related to coping with DMD-related uncertainty and fear. Efforts to improve mothers' wellbeing should focus on interventions to foster mothers' resilience, especially as mothers age, and to enhance benefit finding through identification of positive aspects of living with DBDM on the caregiver and family. Interventions that target perceived burden may not be effective at improving long-term wellbeing. Further research should evaluate a brief needs assessment and an item measuring positive DBMD impact in clinical settings. The use of needs assessment instruments in clinical encounters is well characterized; for example, as described by Boneskvi and colleagues.⁴³ The single impact item offers a simple, positively-valenced assessment of finding benefit in a challenging situation, and use of the item may help identify mothers for whom psychosocial interventions may be targeted. Overall, the results of this study suggest that clinical interventions can address unmet needs while highlighting strengths and

wellbeing, rather than burden and deficit.

CONFLICT OF INTEREST

Associate Professor Bettina Meiser receives a Career Development Award Level 2 from the NHMRC. Holly Peay, Kathleen Kinnett, Pat Furlong, Kathryn Porter, and Aad Tibben declare no potential conflict of interest.

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