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Labor market participation and productivity costs for female caregivers of minor male children with Duchenne and Becker muscular dystrophies

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Abstract

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Rieza H. Soelaeman was the primary analyst and Michael G. Smith independently replicated the analysis. The findings and conclusions in this report are those of the authors and do not necessarily represent the official position of the Centers for Disease Control and Prevention.

AUTHOR CONTRIBUTIONS

Rieza H. Soelaeman led the development of this manuscript, including study conception and design, data analysis and interpretation, and drafting the manuscript. Dana Goodenough, Scott D. Grosse, Lijing Ouyang, Pangaja Paramsothy, Kashika Sahay, Michael G. Smith, and J. Mick Tilford contributed to the conception and design of the study. Scott D. Grosse, Kashika Sahay, and J. Mick Tilford drafted sections of the manuscript. Michael G. Smith independently replicated the analysis. Scott D. Grosse, Joyce Oleszek, Lijing Ouyang, Pangaja Paramsothy, Kashika Sahay, Michael G. Smith, and J. Mick Tilford contributed to the interpretation of the findings. Pangaja Paramsothy and Joyce Oleszek provided extensive feedback throughout manuscript development. Joyce Oleszek ensured the accuracy of clinical discussions. Rieza H. Soelaeman and Scott D. Grosse critically revised the manuscript. All authors approved the final version.

CONFLICT OF INTEREST

None of the authors has any conflict of interest to disclose.

ETHICAL PUBLICATION STATEMENT

We confirm that we have read the Journal's position on issues involved in ethical publication and affirm that this report is consistent with those guidelines.

ETHICS STATEMENT

Local Institutional Review Board approval was obtained at each MD STAR*net* site.

SUPPORTING INFORMATION

Additional supporting information may be found in the online version of the article at the publisher's website.

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Introduction/Aims: Duchenne and Becker muscular dystrophies (DBMD) are X-linked neuromuscular disorders characterized by progressive muscle weakness, leading to decreased mobility and multisystem complications. We estimate productivity costs attributable to time spent by a parent caring for a male child under the age of 18 y with DBMD, with particular focus on female caregivers of boys with Duchenne muscular dystrophy (DMD) who have already lost ambulation.

Methods: Primary caregivers of males with DBMD in the Muscular Dystrophy Surveillance and Research Tracking Network (MD STAR*net*) were surveyed during 2011–2012 on family quality of life measures, including labor market outcomes. Of 211 respondents, 96 female caregivers of boys with DBMD were matched on state, year of survey, respondent's age, child's age, and number of minor children with controls constructed from Current Population Survey extracts. Regression analysis was used to estimate labor market outcomes and productivity costs.

Results: Caregivers of boys with DBMD worked 296 h less per year on average than caregivers of unaffected children, translating to a $8816 earnings loss in 2020 U.S. dollars. Caregivers of boys with DMD with ≥4 y of ambulation loss had a predicted loss in annualized earnings of $23,995, whereas caregivers of boys with DBMD of the same ages who remained ambulatory had no loss of earnings.

Discussion: Female caregivers of non-ambulatory boys with DMD face additional household budget constraints through income loss. Failure to include informal care costs in economic studies could understate the societal cost-effectiveness of strategies for managing DMD that might prolong ambulation.

Keywords

Becker muscular dystrophy; disability; Duchenne muscular dystrophy; informal care; productivity costs

1 | INTRODUCTION

Duchenne and Becker muscular dystrophies (DBMD) are X-linked neuromuscular disorders characterized by progressive muscle weakness that leads to decreased mobility as well as respiratory and cardiac complications.[1] Duchenne muscular dystrophy (DMD) is the more severe phenotype with earlier onset and more rapid progression.[2] Management of DMD involves specialized, time-consuming care; the need for which increases over time.[3] The majority of children and adolescents with special needs, including DBMD, are cared for at home.[4] High-intensity care can lead to parents reporting lower quality of life, increased financial burden, and negative psychological outcomes.[1,5–8]

Although the literature on the psychosocial impacts of caring for individuals with DBMD is fairly robust,[2,5,6,9–17] less is known about the economic cost of informal care, defined as unpaid care provided to individuals with a medical condition or disability.[18] Researchers can use either of two approaches to estimate the "indirect" costs of informal care. In one approach, researchers impute a monetary cost to all reported hours of informal care. For example, one study that used this approach found that informal caregiving costs were the main drivers of the total cost of illness for children age 2–17 y with DMD in seven of

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eight European countries.[12] Two cost studies assessed loss of labor income, and another study reported reductions in employment due to informal caregiving among parents of males with DMD.[2,12,15,16,19] These studies suggest that informal caregiving may vary by severity in DBMD. A 2014 German study on informal caregivers of males with DMD (ages 1–42 y) and Becker muscular dystrophy (BMD, ages 2.5–62 y) found that 89% of patients with DMD and 47% of patients with BMD received care, usually from a parent.[2] The researchers reported that 29% of parents of males with DMD stopped working to care for their child, and among those who worked, 38% reduced hours by an average of 15 h/wk. By contrast, few parents of males with BMD stopped (4%) or reduced (12%) work.[2] In this study, we estimated caregiving time and forgone earnings related to caring for a boy with DBMD in the United States.

2 | METHODS

2.1 | Data

2.1.1 | Caregivers of boys with DBMD—The Muscular Dystrophy Surveillance, Tracking, and Research Network (MD STAR*net*) is a population-based surveillance system established in 2002 by the Centers for Disease Control and Prevention through cooperative agreements with sites in Arizona (AZ), Colorado (CO), Iowa (IA), and western New York State (NY) with a goal of determining the prevalence of DBMD, and of tracking clinical practices and health outcomes.[20] Surveillance activities at these four sites began in 2004, with two additional sites, Georgia (GA) and Hawaii, joining in 2005 and 2008, respectively. Surveillance methodology, including case identification and ascertainment, are described elsewhere.[20,21] The surveillance dataset contains information on individuals with DBMD who were born since January 1, 1982, diagnosed by age 21, resided in the surveillance area, and were identified by December 31, 2011. Medical record abstraction was completed from 2011 through 2012. Public health authorities permitted medical record abstraction for DBMD at all sites. Institutional review board (IRB) approval or exemption was obtained from the respective institutions.

We used data from the MD STAR*net* Family Quality of Life (FQOL) survey, a special project of MD STAR*net*, as the primary source of information of labor market outcomes for caregivers of male children with DBMD. The FQOL survey is a cross-sectional survey of primary caregivers of males with DBMD identified through MD STAR*net* that asked questions about quality of life, social support, stress, and labor market participation among eligible caregivers of people who opted to take the survey.[22,23] The questions on labor market participation were modeled after the U.S. Census Bureau and Bureau of Labor Statistics Current Population Survey (CPS). The FQOL survey was administered in six MD STAR*net* sites between 2011 and 2012. IRB approval was obtained from all sites for the FQOL survey. Except for CO, where written consent was required, respondent consent to participate was implied if they returned the survey. Among 460 caregivers of affected individuals invited to participate in the FQOL survey, 211 (46%) completed the survey. Selection for study inclusion is shown in the Supporting Information Figure S1, which is available online.[23] Ninety-six female caregivers of boys with DBMD (86 DMD and 10 BMD) were included.

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2.1.2 | Caregivers of children in the comparison group—To estimate earnings forgone due to providing care for children with DBMD beyond usual caregiving, we constructed a comparison group using the March Supplement of the CPS from each state for the year in which the FQOL caregiver survey took place (2011 for AZ and CO, and 2012 for CO, GA, IA, and NY).[24–27] We restricted the comparison group to adult females with at least one own child under age 18 y (n = 4988).

2.1.3 | Matching—We matched caregivers in the case and comparison groups on state, year of interview or survey, respondent's age, child's age, and number of children under 18 y in the household. A case-to-control ratio of approximately 1:4 was used to increase statistical power and to ensure that the comparison estimates are robust.[28] The final analytic dataset contained information on 96 caregivers of boys with DBMD and 371 caregivers of children in the comparison group.

2.2 | Variables

2.2.1 | Total hourly compensation—Estimation of labor market productivity costs involves estimating the reduction in work hours from caring for a child with DBMD and translating that estimate into productivity cost by multiplying by hourly compensation or earnings. Because caregivers of children with DBMD may be likely to earn less than the general population, hourly earnings were predicted for all caregivers using the National Bureau of Economic Research data extracts of the Outgoing Rotations Group from the 2011 and 2012 CPS to represent their potential earnings.[24,29–31] Use of potential earnings is conservative as any earnings differences due to caring for a child with DBMD are not incorporated into the productivity cost estimates. We computed hourly earnings by dividing the usual weekly earnings by usual weekly hours worked by the respondent on the CPS. Because cash and wage earnings represent only a fraction of an individual's earning capacity, we adjusted reported hourly earnings to include fringe benefits using information from the 2011 and 2012 Employer Costs for Employee Compensation (ECEC) survey.[32–34] The ECEC survey measures the average cost to employers of wages, salaries, and benefits in the U.S. civilian workforce. We used the same adjustment factor for all combinations of state of residence, age, and sex because fringe benefits data are not available below the national level. Finally, we inflated total compensation to their December 2020 dollar value using the Employment Cost Index.[35] We took the mean of the adjusted total hourly compensation for each survey year, state of residence, age, and sex combination, and used it to convert hours of labor productivity lost to a monetary value. We assumed self-employed persons had the same earning capacity as wage and salary workers.[34]

2.2.2 | Disease severity—We expected productivity costs to differ by disease severity.[2] We used the number of years since ambulation loss as of the survey date as a proxy of severity of illness among boys with DBMD and categorized it as follows: no ambulation loss, 0–3 y of ambulation loss, and four or more years of ambulation loss. In our sample, all boys with ambulation loss (n = 44) had DMD, whereas those without ambulation loss could have either DMD (n = 42) or BMD (n = 10). The associations between clinical variables indicative of more severe disease and the proxy disease severity variable used in the analysis are shown in Supporting Information Table S1.

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2.3 | Model

All parameters were estimated using regression models. A detailed explanation of the models is presented in the Supporting Information File.

2.4 | Labor market productivity cost calculation

To calculate the labor market productivity costs for caregivers of boys with DBMD, we compared the mean differences in weekly and annualized work hours between caregivers of boys with DBMD and caregivers of children in the comparison group. We converted annualized hours lost to dollar values of earnings lost by multiplying them by the mean predicted total hourly compensation. Finally, we compared the differences in annualized earnings between caregivers of boys with DBMD and caregivers of children in the comparison group, in aggregate and by severity of illness, using a t-test. In all analyses, we clustered the SE around the case–control pair identifier to reflect the case–control matching. All data management and statistical analyses were performed in Stata 14 SE (StataCorp, College Station, TX, USA).

3 | RESULTS

3.1 | Participant characteristics

Caregiver age ranged from 38.0 to 44.9 y, with a higher percentage of caregivers of boys with DBMD who were non-Hispanic white than caregivers in the comparison group (Table 1). Both sets of caregivers were similar in their levels of employment and numbers of weeks worked, but caregivers of boys with DBMD worked fewer hours per week. Caregivers of boys with DBMD and ambulation loss were also less likely to have worked in the past year than caregivers of boys who were ambulatory, while caregivers of boys with four or more years of ambulation loss worked the fewest number of weeks.

3.2 | Multivariable regression results

Caregivers of boys with four or more years of ambulation loss were 29 percentage points less likely than caregivers of children in the comparison group to have worked in the past year (Table 2, Column A; odds ratios provided in Supporting Information Table S2). Caregiver age was positively associated with having worked in the past year, although this effect diminished at older ages. Employed caregivers of boys with DBMD did not differ from caregivers of children in the comparison group in the number of weeks worked in the past year (Table 2, Column B). Finally, employed caregivers of boys with DMD and ambulation loss worked fewer hours per week than caregivers in the comparison group with the greatest reductions among caregivers of boys with four or more years of ambulation loss (Table 2, Column C). Child age was positively associated with the number of hours worked per week, with each additional year of age corresponding to a 0.8 h increase in working time.

Regression models that included only caregivers of affected males (children and adults) confirm the importance of ambulation loss as a predictor of labor market outcomes (Supporting Information Table S3). Notably, caregivers of persons who lost ambulation at ages 6–8 y were 48 percentage points less likely to work than caregivers of those without

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ambulation loss. Simulated caregiver weekly work hours for a hypothetical 10-y-old boy were highest if he were ambulatory and lowest if he lost ambulation between ages 6–8 y (Supporting Information Table S4). For a hypothetical affected person older than age 10 y, simulated caregiver weekly work hours were lowest if ambulation were lost between ages 6–8 y and highest if ambulation were lost at age 11 y.

3.3 | Labor market productivity costs

Compared to caregivers of children in the comparison group, caregivers of boys with DBMD worked 295.9 h less per year, corresponding to a $8816 loss in predicted earnings (base wages and salaries plus fringe benefits) in 2020 U.S. dollars (Table 3). When disaggregated by disease severity, losses were observed for caregivers of boys with ambulation loss compared to caregivers in the comparison group, but not for caregivers of boys with DBMD who were ambulatory. Predicted losses increased with longer duration of ambulation loss: caregivers of boys with four or more years of ambulation loss faced the highest labor market productivity costs with an annualized earnings loss of $23,995 compared to $13,828 for caregivers of boys with 0–3 y of ambulation loss.

4 | DISCUSSION

We found that overall, the percentage of caregivers of boys with DBMD employed in the past year was only slightly lower compared to caregivers in the comparison group, 67.7% versus 72.0%. By contrast, several previous studies noted that many parents of boys with DMD reported having reduced employment or hours of work because of their child's condition.[2,12,15] In one of the studies, researchers interviewed 770 caregivers in four countries (Germany, Italy, United Kingdom, and United States) and found that 27%–49% reported having reduced working hours or stopped working completely due to DMD.[14] The share was lowest among caregivers in the United States, possibly because unlike European countries, access to health insurance is usually conditioned on continued employment, which is a disincentive for caregivers to stop working completely. More than half of parents of children with muscular dystrophy in a U.S. survey reported having stopped working or reduced hours of work due to their child's condition, a much higher proportion than for parents of children with other special healthcare needs.[36] However, none of those studies compared employment patterns with those of demographically matched parents of unaffected children.

More importantly, we demonstrated that caregivers of boys with four or more years of ambulation loss were significantly less likely to be employed than caregivers of children in the comparison group. This finding differs from that of a German study which reported that caregivers of boys with DMD who stopped working mostly did so when their child was still ambulatory but was having difficulty with ambulation. It should be noted that, even though we observed higher productivity loss among caregivers of boys who had lost ambulation the longest, we cannot conclude that longer ambulation loss necessarily leads to greater productivity loss. That is because the duration of ambulation loss is correlated with child age and disease severity; older boys with DBMD are both more likely to be non-ambulatory and to have other disease complications.

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We found an offsetting independent positive association of age among boys with DBMD with caregiver labor market outcomes and no significant interaction between child age and case status (Table 2). Nevertheless, the inclusion of caregiver time or productivity costs in economic studies could more fully account for the costs of DBMD from the societal perspective. These estimates could be used in economic evaluations of strategies to help prevent, treat, or manage DBMD. Our results corroborate the findings of previous studies that families of people with DMD face substantial indirect medical and non-medical costs, including income loss.[1,2,5,6,9–17,19,37–41]

The labor market productivity cost estimates from the multivariate models were especially revealing. The $23,995 reduction in predicted annual earnings for caregivers of boys with DMD who had been non-ambulatory the longest is equivalent to 41% of the 2011–2012 median household income in the United States.[42] It is similar in magnitude to Larkindale et al.'s report of $25,670 lower household earned income for households having a child with neuromuscular disease, including DMD, where that child required 16–24 h of care relative to households where no informal care was required.[19] The authors also reported that households with children with neuromuscular disease who required less care experienced smaller, non-significant reductions in earned income.[19]

The findings in this study are also similar in some respects to previous work examining the labor productivity costs for caregivers of children with spina bifida.[31] Caregivers of children with spina bifida had a 21–27 percentage point lower probability of working in the past year compared to a 16–17 percentage point lower probability for caregivers of boys with DMD and ambulation loss. While there were significantly fewer weeks worked for caregivers of children with spina bifida, there was little difference in hours worked per week. In contrast, caregivers of boys with DMD and ambulation loss had significantly fewer hours worked per week, but no statistical difference in weeks worked. Overall, the mean predicted reduction in weekly work hours were similar across the two conditions, with confidence intervals overlapping for caregivers of boys with DMD and ambulation loss (−10.7 to −15.0) (Table 3) compared to caregivers of children with spina bifida and higher impairment level (−9.8 to −11.3).[31]

In the natural history of DMD, ambulation loss occurs at a median age of 12 y followed by rapid progression of orthopedic comorbidities such as muscle contractures and scoliosis.[16] As function declines and complexity of medical care increases, more caregiver assistance is needed. With disease progression and ambulation cessation, decline in pulmonary function results in the need for more respiratory interventions such as nighttime mechanical ventilation. Similarly, advancing cardiomyopathy requires more frequent cardiac monitoring, and scoliosis is more likely to require surgery.

One major limitation of this study is the use of the opportunity cost approach to estimate informal caregiving costs as the reduction in paid employment instead of directly measuring and placing a monetary value on all hours of caregiving time. Lost earnings due to the displacement of paid work by informal care responsibilities underestimates informal caregiving costs because it implicitly assigns a zero value to care that does not substitute for paid employment.[18]

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In addition, survey respondents were a small, all-female subset (46%) of eligible caregivers in MD STAR*net*, and the results may not be generalizable to caregivers of U.S. males living with DBMD. A previous analysis found that FQOL survey respondents were more likely to be non-Hispanic white and to live in wealthier census tracts than non-respondents, which may further limit generalizability of the findings.[23] There may also have been unobserved differences. For example, individuals in dual-income households may be more likely to respond to the survey or to forego work to care for their child with DBMD.

An inherent limitation of observational data is the inability to establish causal effects from cross-sectional associations. Since one cannot assign families to have a child with a disorder, it is impossible to prove that having a child with DMD reduces caregiver employment. However, it is established practice for researchers to estimate caregiver productivity costs using observational data.

Our results present a partial estimate of the economic impact of caring for a child with DBMD on caregivers' employment and earnings. Our estimates of productivity costs, which assume that these caregivers have the same hourly earnings as the comparison group, are based on reductions in annual hours of work and do not quantify the impact of DMD caregiving responsibilities on earnings per hour worked. The median hourly earnings were lower in the FQOL sample than in the matched CPS sample, $22.56 versus $28.75. The lower hourly earnings for employed caregivers of children with DMD likely reflect the cumulative impact of interruptions in employment or restrictions on hours of work due to increased caregiving demands. Studies that report differences in labor income, which is the product of annual hours of work and mean hourly earnings, can yield larger estimates of productivity costs.[18]

In conclusion, caregivers of children with DMD are often faced with high costs related to the disease, including direct medical costs and non-medical costs as well as caregiver time costs. In this study, we estimated that the labor market productivity losses for female caregivers of boys with DMD and ambulation loss were conservatively $10,000–25,000 higher relative to caregivers of children in the comparison group. We suggest that inclusion of caregiver labor market productivity or informal care time costs in economic evaluations and cost-of-illness studies that are conducted from the societal perspective could lead to more accurate assessments of the cost-effectiveness of interventions and strategies for managing DBMD.

Supplementary Material

Refer to Web version on PubMed Central for supplementary material.

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DATA AVAILABILITY STATEMENT

Due to privacy concerns data from the MD STARnet is not publicly available. Data used for this analysis are maintained at the Centers for Disease Control and Prevention. Researchers interested in MD STARnet should contact MD STARnet at MDSTARnet@cdc.gov.

ABBREVIATIONS:

AZ	Arizona
BMD	Becker muscular dystrophy
CO	Colorado
CPS	Current Population Survey
DBMD	Duchenne and Becker muscular dystrophies
DMD	Duchenne muscular dystrophy
ECEC	Employer Costs for Employee Compensation
FQOL	family quality of life
IA	Iowa
IRB	institutional review board
MD STAR*net*	Muscular Dystrophy Surveillance, Tracking, and Research Network
NY	New York

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TABLE 1

Characteristics of female caregivers and their children

Characteristic	Severity of illness[a]			All caregivers of boys with DBMD (n = 96)	Control-CPS caregivers (n = 371)	*P*-Value*
	No ambulation loss (n = 52)	0–3 y of ambulation loss (n = 19)	4+ y of ambulation loss (n = 25)			
Caregiver age (y), mean (SE)	38.0 (0.98)	42.3 (1.61)	44.9 (1.56)	40.6 (0.80)	40.1 (0.72)	.190
Caregiver race/ethnicity, n (%)						
Non-Hispanic white	45 (86.5)	15 (79.0)	17 (68.0)	77 (80.2)	211 (56.9)	<.001
Other	7 (13.5)	4 (21.1)	8 (32.0)	19 (19.8)	160 (43.1)	
Caregiver education, n (%)[b]						
Less than high school	5 (9.6)	4 (21.1)	2 (8.3)	11 (11.6)	51 (13.8)	.08
High school graduate	9 (17.3)	3 (15.8)	3 (12.5)	15 (15.8)	100 (27.0)	
Some college or trade	19 (36.5)	5 (26.3)	12 (50.0)	36 (37.9)	109 (29.4)	
College graduate	19 (36.5)	7 (36.8)	7 (29.2)	33 (34.7)	111 (29.9)	
Caregiver marital status, n (%)						
Married	46 (88.5)	15 (79.0)	16 (64.0)	77 (80.2)	287 (77.4)	.33
Divorced	5 (9.6)	3 (15.8)	5 (20.0)	13 (13.5)	44 (11.9)	
Single, never married	1 (1.9)	1 (5.3)	4 (16.0)	6 (6.3)	40 (10.8)	
Multiple affected children in the household	3 (5.8)	0 (0.0)	2 (8.0)	5 (5.2)	0 (0.0)	N/A
Number of children <18 in household, mean (SE)	2.1 (0.2)	2.1 (0.2)	1.5 (0.1)	2.0 (0.1)	2.0 (0.1)	.06
Presence of children <6 in the household, n (%)	17 (32.7)	3 (15.8)	0 (0.0)	20 (20.8)	97 (26.2)	.21
Age of matched child, mean (SE)	9.4 (0.5)	13.1 (0.5)	15.4 (0.3)	11.7 (0.4)	11.6 (0.4)	.09
Labor market outcomes[c]						
Worked in the past year, n (%)	39 (75.0)	10 (58.8)	14 (58.3)	63 (67.7)	267 (72.0)	.38
Weeks worked in the past year, mean (SE)[d]	47.3 (1.6)	48.7 (1.9)	40.6 (3.8)	45.8 (1.5)	46.4 (0.7)	.74
Hours worked per week, mean (SE)[d]	33.2 (2.4)	28.9 (4.0)	26.8 (3.7)	31.1 (1.8)	37.3 (0.7)	.002

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[a] "No ambulation loss" includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD.

[b] Caregiver education missing from two caregivers of boys with DBMD.

[c] Labor market outcomes missing from three caregivers of boys with DBMD.

[d] Conditional on having worked in the past year.

[*] Wald F test *P*-values are reported for continuous variables, while Pearson chi-squared test *P*-values are reported for categorical variables. *P*-values are shown for tests of overall difference between the case and comparison groups.

Muscle Nerve. Author manuscript; available in PMC 2022 December 01.

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TABLE 2

Results from multiple regression models of labor market outcomes

	(A) Logistic regression		(B) Linear regression		(C) Linear regression	
	Worked in the past year		Weeks worked		Hours worked per week	
Characteristic	**Marginal effect[a]**	**95% confidence interval**	**Beta**	**95% confidence interval**	**Beta**	**95% confidence interval**
Severity[b]						
Control	Ref.		Ref.		Ref.	
No ambulation loss	0.035	(−0.10, 0.17)	0.39	(−3.38, 4.15)	−3.10	(−8.38, 2.18)
0–3 y of ambulation loss	−0.20	(−0.45, 0.05)	1.03	(−4.65, 6.70)	−8.07	(−15.97, −0.18)
4+ y of ambulation loss	−0.29	(−0.52, −0.05)	−7.93	(−16.39, 0.53)	−12.6	(−21.5, −3.7)
Age of child	0.008	(−0.004, 0.02)	−0.03	(−0.59, 0.53)	0.76	(0.38, 1.15)
Case status * Age of child	0.005	(−0.03, 0.04)	−0.35	(−1.24, 0.54)	−0.02	(−1.13, 1.09)
Caregiver age	0.06	(0.03, 0.10)	0.93	(−0.68, 2.54)	0.89	(−1.25, 3.03)
Caregiver age-squared	−0.001	(−0.001, −0.0004)	−0.009	(−0.03, 0.01)	−0.01	(−0.04, 0.01)
Caregiver race/ethnicity: Non-Hispanic white	0.03	(−0.05, 0.12)	−1.09	(−3.92, 1.74)	−2.28	(−5.04, 0.49)
Caregiver education						
Less than high school	Ref.		Ref.		Ref.	
High school graduate	0.17	(0.04, 0.29)	−4.17	(−9.68, 1.34)	6.71	(2.77, 10.65)
Some college or trade	0.29	(0.16, 0.41)	−1.84	(−7.22, 3.54)	7.92	(3.26, 12.58)
College graduate	0.30	(0.17, 0.43)	−3.35	(−9.07, 2.38)	6.21	(1.75, 10.66)
Married	−0.08	(−0.18, 0.02)	0.40	(−2.49, 3.30)	−0.43	(−2.98, 2.12)
Number of children <18	−0.03	(−0.07, 0.02)	−1.80	(−3.38, −0.22)	0.17	(−1.18, 1.52)
Presence of children <6	−0.04	(−0.16, 0.08)	2.14	(−1.00, 5.29)	3.14	(−0.08, 6.36)
Child had a behavioral or mental health diagnosis	−0.10	(−0.18, −0.01)	−1.56	(−4.14, 1.03)	0.85	(−1.36, 3.06)
Child diagnosed with cardiomyopathy	0.08	(−0.19, 0.34)	6.69	(−0.62, 13.99)	0.79	(−7.98, 9.56)

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	(A) Logistic regression		(B) Linear regression		(C) Linear regression	
	Worked in the past year		Weeks worked		Hours worked per week	
Characteristic	**Marginal effect[a]**	**95% confidence interval**	**Beta**	**95% confidence interval**	**Beta**	**95% confidence interval**
Multiple affected children in household	−0.09	(−0.36, 0.18)	1.93	(−3.89, 7.75)	0.20	(−7.60, 8.00)
Constant			30.22	(−5.60, 66.04)	16.82	(−28.19, 61.82)
R^{2}[c]	0.14		0.06		0.13	
N	463		323		326	

[a] Marginal effect: Average change in the predicted probability of the event occurring (adjusted risk difference) corresponding to a change in the regressor.

[b] "No ambulation loss" includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD.

[c] Pseudo R^2 is shown for the logistic regression model and adjusted R^2 is shown for the linear regression models.

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TABLE 3

Average reduction in hours worked per week, hours worked per year, and annualized labor market productivity costs for caregivers of boys with DBMD

Comparison with caregivers of children in the comparison group	Mean loss in work hours per week (95% confidence interval)	Mean loss in annualized work hours (95% confidence interval)	Total annual productivity costs (2020 U.S. dollars)[a]		
			Mean	Lower limit	Upper limit
Overall	−6.4 (−8.2, −4.6)	−295.9 (−384.6, −207.3)	−$8816	−$11,604	−$6027
Severity[b]					
No ambulation loss	−1.0 (−3.2, 1.3)	−1.5 (−111.1, 108)	−$41	−$3593	$3511
0–3 y of ambulation loss	−10.7 (−14.8, −6.7)	−459.0 (−658.2, −259.8)	−$13,828	−$20,124	−$7533
4+ y of ambulation loss	−15.0 (−18.5, −11.5)	−809.4 (−977, −641.8)	−$23,995	−$29,058	−$18,931

[a]Mean earnings by age and sex for the state and year of survey using Current Population Survey data were used to value loss in annualized work hours.

[b]"No ambulation loss" includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD.