**Supplemental File**

**Supplemental Methods**

*Regression models*

The probability of having worked in the past 12 months was estimated using logistic regression on the entire analytic sample, whereas the number of weeks worked in a year and hours worked per week were estimated using linear regression on the sub-sample that worked within one year of the survey. The severity of illness was entered as three dummy variables: DBMD with no ambulation loss; DMD with 0-3 years of ambulation loss; and DMD with 4 or more years of ambulation loss. Other independent variablesincluded caregiver’s age, race, education, marital status, number of children under 18 years, presence of children under 6 years, whether there were multiple children with DBMD in the household, and age of the affected child standardized relative to the sample mean. The number of minor children in the household was derived from the MD STAR*net* supplemental Parent Interview, while the number of children with DBMD came from the surveillance dataset. To control for the potential confounding of the disease severity measure by child age for boys with DBMD, we added a case status\*standardized child age interaction term in all regression models. We estimated the predicted probability of working in the past year, number of weeks worked last year, and hours worked per week for all caregivers in the dataset. We used the parameter estimates from the models, conditional on having worked within one year of the survey, to make out-of-sample predictions of weeks worked in a year and hours worked per week for those who did not work in the past year.

Because an affected individual’s care needs increase over time, we examined factors associated with the three labor market outcomes for adult and minor males with DBMD separately using age at ambulation loss as a model covariate instead of the disease severity proxy variable, and simulated caregiver weekly work hours for a hypothetical male with DBMD at several combinations of age at ambulation loss and current age of the affected person and present these results in **Supplemental Tables 3 and 4**. We kept the following parameters constant in the simulation: non-Hispanic white male with no diagnosis of behavioral disorder, no diagnosis of cardiomyopathy, whose mother is married with some college education and 25 years old at son's birth. There are no children under 6, no other children under 18, and no other children with DBMD in the household. We simulated caregiver weekly work hours for affected individuals at ages 10,12, 14, 16, 18, and 20 years old.

**Supplemental Tables and Figure**

**Supplemental Table 1.** Association between the disease severity proxy variable and select clinical outcome variables

|  |  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Clinical characteristic** | **Severity**\* | | | | | | **Total (n=96)** | | **p-value**† |
| **No ambulation loss (n=52)** | | **0-3 years of ambulation loss  (n=19)** | | **4+ years of ambulation loss (n=25)** | |
| Parent reported that child experienced loss of arm function |  |  |  |  |  |  |  |  |  |
| No | 51 | (98.1) | 14 | (73.7) | 2 | (8.0) | 67 | (69.8) | <0.001 |
| Yes | 1 | (1.9) | 5 | (26.3) | 23 | (92.0) | 29 | (30.2) |  |
| Parent reported that child experienced loss of leg function |  |  |  |  |  |  |  |  |  |
| No | 48 | (92.3) | 0 | (0.0) | 0 | (0.0) | 48 | (50.0) | <0.001 |
| Yes | 4 | (7.7) | 19 | (100.0) | 25 | (100.0) | 48 | (50.0) |  |
| Child had scoliosis surgery |  |  |  |  |  |  |  |  |  |
| No | 52 | (100.0) | 17 | (89.5) | 11 | (44.0) | 80 | (83.3) | <0.001 |
| Yes | 0 | (0.0) | 2 | (10.5) | 14 | (56.0) | 16 | (16.7) |  |
|  |  |  |  |  |  |  |  |  |  |
|  |  |  |  |  |  |  |  |  |  |
| **Clinical characteristic** | **Severity**\* | | | | | | **Total (n=96)** | | **p-value**† |
| **No ambulation loss (n=52)** | | **0-3 years of ambulation loss  (n=19)** | | **4+ years of ambulation loss (n=25)** | |
| Child uses non-invasive assisted ventilation |  |  |  |  |  |  |  |  |  |
| No | 51 | (98.1) | 18 | (94.7) | 12 | (48.0) | 81 | (84.4) | <0.001 |
| Yes | 1 | (1.9) | 1 | (5.3) | 13 | (52.0) | 15 | (15.6) |  |
| Child had a cardiomyopathy diagnosis |  |  |  |  |  |  |  |  |  |
| No | 46 | (88.5) | 14 | (73.7) | 20 | (80.0) | 80 | (83.3) | 0.29 |
| Yes | 6 | (11.5) | 5 | (26.3) | 5 | (20.0) | 16 | (16.7) |  |
| Child had a behavioral or mental health diagnosis |  |  |  |  |  |  |  |  |  |
| No | 32 | (61.5) | 10 | (52.6) | 11 | (44.0) | 53 | (55.2) | 0.34 |
| Yes | 20 | (38.5) | 9 | (47.4) | 14 | (56.0) | 43 | (44.8) |  |

Abbreviations: DMD = Duchenne muscular dystrophy; BMD = Becker muscular dystrophy

\*“No ambulation loss” includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD

†Pearson’s chi-squared test p-values shown for tests of overall difference across disease severity categories

**Supplemental Table 2.** Adjusted odds ratios (OR) from multiple logistic regression model of the likelihood of working in the past 12 months

|  |  |  |
| --- | --- | --- |
| **Characteristic** | **Adjusted odds ratio** | **95% confidence interval** |
| Severity\* |  |  |
| Control | Ref. | Ref. |
| No ambulation loss | 1.24 | (0.50, 3.10) |
| 0-3 years of ambulation loss | 0.35 | (0.11, 1.16) |
| 4+ years of ambulation loss | 0.24 | (0.07, 0.76) |
| Age of child | 1.05 | (0.97, 1.14) |
| Case status \* Age of child | 1.03 | (0.84, 1.26) |
| Caregiver age | 1.45 | (1.17, 1.80) |
| Caregiver age-squared | 1.00 | (0.99, 1.00) |
| Caregiver race/ethnicity: non-Hispanic white | 1.22 | (0.73, 2.04) |
| Caregiver education |  |  |
| Less than high school | Ref. | Ref. |
| High school graduate | 2.68 | (1.24, 5.77) |
|  |  |  |
|  |  |  |
| **Characteristic** | **Adjusted odds ratio** | **95% confidence interval** |
| Some college or trade | 5.41 | (2.37, 12.38) |
| College graduate | 5.73 | (2.53, 12.97) |
| Married | 0.62 | (0.34, 1.12) |
| Number of children <18 | 0.86 | (0.65, 1.12) |
| Presence of children <6 | 0.80 | (0.38, 1.66) |
| Child had a behavioral or mental health diagnosis | 0.57 | (0.35, 0.94) |
| Child diagnosed with cardiomyopathy | 1.56 | (0.32, 7.63) |
| Multiple affected children in household | 0.59 | (0.12, 2.99) |

Abbreviations: DMD = Duchenne muscular dystrophy; BMD = Becker muscular dystrophy

\*“No ambulation loss” includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD

**Supplemental Table 3.** Factors associated with labor market outcomes among caregivers of adult and minor males with DBMD (n=169)

|  |  |  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- | --- | --- |
| **Characteristic** | **(A) Logistic regression** | |  | **(B) Linear regression** | |  | **(C) Linear regression** | |
| **Worked in the past year** | |  | **Weeks worked** | |  | **Hours worked per week** | |
| **Marginal Effect\*** | **95% confidence interval** |  | **Beta** | **95% confidence interval** |  | **Beta** | **95% confidence interval** |
| Age at ambulation loss† |  |  |  |  |  |  |  |  |
| No ambulation loss | Ref. |  |  | Ref. |  |  | Ref. |  |
| 6-8 years old | -0.48 | (-0.71, -0.26) |  | -6.13 | (-16.7, 4.5) |  | -1.77 | (-13.3, 9.8) |
| 9 years old | -0.16 | (-0.41, 0.09) |  | -6.72 | (-16.7, 3.2) |  | -2.32 | (-12.5, 7.9) |
| 10 years old | -0.26 | (-0.49, -0.03) |  | -12.9 | (-22.2, -3.6) |  | -5.35 | (-15.1, 4.4) |
| 11 years old | 0.03 | (-0.15, 0.22) |  | -1.11 | (-10.4, 8.2) |  | 2.21 | (-8.0, 12.4) |
| 12 years old | -0.35 | (-0.64, -0.06) |  | -5.34 | (-16.5, 5.8) |  | -5.70 | (-16.8, 5.4) |
| 13+ years old | -0.36 | (-0.6, -0.12) |  | 1.36 | (-8.0, 10.8) |  | 3.06 | (-7.2, 13.3) |
| Age of affected person | 0.01 | (-0.01, 0.03) |  | -0.64 | (-1.4, 0.2) |  | 0.73 | (-0.1, 1.6) |
|  |  |  |  |  |  |  |  |  |
| **Characteristic** | **(A) Logistic regression** | |  | **(B) Linear regression** | |  | **(C) Linear regression** | |
| **Worked in the past year** | |  | **Weeks worked** | |  | **Hours worked per week** | |
| **Marginal Effect**\* | **95% confidence interval** |  | **Beta** | **95% confidence interval** |  | **Beta** | **95% confidence interval** |
| Caregiver age | 0.07 | (0.01, 0.14) |  | 0.61 | (-2.2, 3.4) |  | 0.66 | (-2.4, 3.7) |
| Caregiver age-squared | -0.001 | (-0.001, -0.0002) |  | 0.00 | (-0.03, 0.02) |  | -0.01 | (-0.04, 0.03) |
| Caregiver race/ethnicity: non-Hispanic white | 0.06 | (-0.12, 0.24) |  | 0.36 | (-7.9, 8.6) |  | 3.15 | (-5.1, 11.4) |
| Caregiver Education |  |  |  |  |  |  |  |  |
| Less than high school | Ref. |  |  |  |  |  |  |  |
| High school graduate | 0.35 | (0.09, 0.61) |  | -7.12 | (-24.2, 10) |  | 13.26 | (-3.3, 29.9) |
| Some college or trade | 0.35 | (0.11, 0.6) |  | -2.82 | (-19.1, 13.5) |  | 14.31 | (-1.5, 30.1) |
| College graduate | 0.34 | (0.09, 0.6) |  | -4.85 | (-21.5, 11.8) |  | 13.63 | (-2.6, 29.9) |
| Married | -0.05 | (-0.21, 0.1) |  | -3.40 | (-10.1, 3.3) |  | -5.00 | (-12.2, 2.2) |
| Number of children <18 | -0.01 | (-0.08, 0.05) |  | -1.44 | (-5.0, 2.1) |  | -0.03 | (-3.9, 3.9) |
| Presence of children <6 | -0.06 | (-0.29, 0.16) |  | 4.05 | (-6.2, 14.3) |  | 14.99 | (3.8, 26.2) |
|  |  |  |  |  |  |  |  |  |
| **Characteristic** | **(A) Logistic regression** | |  | **(B) Linear regression** | |  | **(C) Linear regression** | |
| **Worked in the past year** | |  | **Weeks worked** | |  | **Hours worked per week** | |
| **Marginal Effect**\* | **95% confidence interval** |  | **Beta** | **95% confidence interval** |  | **Beta** | **95% confidence interval** |
| Affected person had a behavioral or mental health diagnosis | -0.02 | (-0.15, 0.11) |  | -0.57 | (-5.9, 4.8) |  | -2.40 | (-8.0, 3.2) |
| Affected person diagnosed with cardiomyopathy | 0.07 | (-0.08, 0.23) |  | 6.48 | (-0.2, 13.1) |  | 0.06 | (-6.6, 6.7) |
| Multiple affected people in household | -0.07 | (-0.28, 0.14) |  | 2.85 | (-5.8, 11.5) |  | -1.27 | (-10.9, 8.4) |
| Constant |  |  |  | 46.3 | (-23.1, 115.8) |  | -6.06 | (-81.2, 69.1) |
| R2‡ | 0.23 |  |  | 0.06 |  |  | 0.04 |  |
| *N* | 169 |  |  | 115 |  |  | 118 |  |

Abbreviations: DBMD = Duchenne and Becker muscular dystrophies; DMD = Duchenne muscular dystrophy; BMD = Becker muscular dystrophy

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

† “No ambulation loss” includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD

‡ Pseudo R2 is shown for the logistic regression model and adjusted R2 is shown for the linear regression models

**Supplemental Table 4**. Simulated number of hours worked per week for caregivers of adult and minor males with DBMD based on regression modeling results using age at ambulation loss and current age of affected person for predictions (n=169)

|  |  |  |  |  |  |  |
| --- | --- | --- | --- | --- | --- | --- |
| **Age at ambulation loss\*** | **Current age of affected person with DBMD** | | | | | |
| **10 years old** | **12 years old** | **14 years old** | **16 years old** | **18 years old** | **20 years old** |
| No ambulation loss | 7.6 | 11.1 | 15.4 | 20.2 | 25.2 | 29.9 |
| 6-8 years old | 2.4 | 4.5 | 7.1 | 10.6 | 15.2 | 20.3 |
| 9 years old | 4.8 | 7.4 | 10.9 | 15.2 | 20.2 | 25.1 |
| 10 years old | 3.3 | 5.5 | 8.4 | 12.1 | 16.8 | 21.5 |
| 11 years old | Not estimated | 13.2 | 17.9 | 22.8 | 27.9 | 32.5 |
| 12 years old | Not estimated | 4.7 | 7.3 | 10.7 | 15.2 | 20.0 |
| 13+ years old | Not estimated | Not estimated | 9.9 | 14.2 | 19.7 | 25.3 |

Abbreviations: DBMD = Duchenne and Becker muscular dystrophies; DMD = Duchenne muscular dystrophy; BMD = Becker muscular dystrophy

\*“No ambulation loss” includes individuals with DMD and BMD. Non-ambulatory categories include individuals with DMD

**Supplemental Figure.** Selection of female caregivers of boys with DBMD

Abbreviations: DBMD = Duchenne and Becker muscular dystrophies