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## **ABBREVIATION**

NSCH

National Survey of Children's Health

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**AIM** To examine how social factors might mitigate the elevated risk of mental health disorders in children with cerebral palsy (CP).

**METHOD** This cross-sectional study included 6- to 17-year-olds with (n=111; 40.4% 6–11y, 59.6% 12–17y) and without (n=29 909; 50.2% 6–11y, 49.8% 12–17y) CP from the 2016 National Survey of Children's Health. Mental health disorders included depression, anxiety, behavior/conduct problems, and attention-deficit/hyperactivity disorder. Social factors included participation in activities, bully victimization, and difficulty with friendships.

**RESULTS** After adjusting for sociodemographic factors and the presence of chronic pain, children with CP had higher odds of anxiety (odds ratio [OR] 4.4; 95% confidence interval [CI] 1.9–8.5), behavior/conduct problems (OR 3.9; 95% CI 1.4–11.3), and multimorbidity (OR 2.8; 95% CI 1.1–7.0), but not depression (OR 1.4; 95% CI 0.6–3.8) or attention-deficit/hyperactivity disorder (OR 1.7; 95% CI 0.6–4.6), compared to controls. With adjustment for participation in activities, the odds of anxiety, behavior/conduct problems, and multimorbidity remained increased in children with CP. With adjustment for difficulty with friendships, the odds of anxiety, behavior/conduct problems, and multimorbidity were no longer increased in children with CP. With adjustment for bully victimization, the odds of behavior/conduct problems and multimorbidity were attenuated in children with CP; however, the odds of anxiety remained increased.

**INTERPRETATION** The elevated prevalence of certain mental health disorders in children with CP is partly associated with modifiable social factors.

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[Right page footer] Mental Health in Children with Cerebral Palsy *Daniel G Whitney et al.* 

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#### What this paper adds

- Difficulty with friendships predicts an elevated prevalence of psychiatric conditions in children with cerebral palsy (CP).
- Bully victimization predicts an elevated prevalence of behavior/conduct problems in children with CP.
- Low participation does not predict mental health disorders in this population.

## [main text]

Cerebral palsy (CP) refers to a group of motor disorders with a considerable range of motor dysfunction. It is the most common pediatric physical disability.<sup>1</sup> Individuals with CP are at a heightened risk for mental health disorders with prevalence rates up to 57%.<sup>2,3</sup> This is concerning because mental health disorders are among the leading causes of the overall global burden of disease among children and adolescents.<sup>4</sup> Further, psychopathology screening as part of a multidisciplinary follow-up in children and adolescents with CP has been recommended.<sup>3</sup> Therefore, it is important to identify the risk factors associated with mental health disorders in children with CP.

There is evidence that physical problems and speech difficulties that are inherent to CP (e.g. motor dysfunction, dysarthria),<sup>5,6</sup> in addition to medical comorbidities (e.g. sleep disorders),<sup>7</sup> can affect mental health. Physical<sup>8–10</sup> and communication<sup>5</sup> impairments can alter the individual's interaction with the physical and social environment, thus affecting social development. Furthermore, social factors can have a profound impact on the individual's mental health status, such as quality of life and self-concept.<sup>10</sup>

Children with CP have less participation and enjoyment of social and recreational activities compared to typically developing children.<sup>11,12</sup> Specifically, they spend less time with<sup>12</sup> and perceive lower social support from<sup>13</sup> friends compared to the general adolescent population, providing a barrier to social development. Despite these social difficulties, children with CP place a high value and need for social and emotional networks,<sup>14–16</sup> such as friendships.<sup>15</sup> Friendships play an important role in the subjective well-being of children and have been shown to decrease the risk of developing mental health disorders.<sup>17</sup> Evidence suggests that friendships may serve as a buffer to lessening the socially harmful effects of bullying,<sup>18</sup> which is important because children with CP are at a heightened risk for being bullied.<sup>19–21</sup> In the general population

of adolescents, victims of bullying have a myriad of risk factors pertaining to poor mental health<sup>22,23</sup> compared to non-victimized adolescents.

Mental health disorders are among the leading causes of the overall pediatric global burden of disease.<sup>4</sup> Thus, it is particularly important to examine mental health and associated risk factors in individuals with CP during the earlier phases of development. Accordingly, the purpose of this study was to examine how social factors, including participation in activities, difficulty with friendships, and bully victimization, in children with CP associate with several mental health disorders in this pediatric population. We hypothesized that each social factor would individually account for some of the elevated prevalence of mental health disorders in children with CP.

## METHOD

The 2016 National Survey of Children's Health (NSCH) was conducted from June 2016 to February 2017 and was designed to generate state-level and national-level data on the health status of American children up to 17 years of age, their families, and their communities. Surveys were completed by mail and using online data collection instruments with a two-phase self-administered data collection design. Over 50 000 interviews of households that had children 0 to 17 years of age were completed across the USA. As previously described,<sup>24</sup> one eligible child per household was randomly selected for the detailed questionnaire and children with special healthcare needs were oversampled to allow for more robust data estimates. The respondent was an adult (usually a parent) who had knowledge of the health status of the surveyed child. The overall weighted response rate was 40.7%. The completion rate for households that initiated the survey was 69.7%. The United States Census Bureau conducted a nonresponse bias analysis for the 2016 NSCH<sup>25</sup> and concluded that, although response was higher in geographical locations where income was higher, there is no strong or consistent evidence of non-response bias. Additional information about the NSCH methodology is available from the Data Resource Center for Child & Adolescent Health (http://www.childhealthdata.org/).

#### Participants

Details regarding participant selection have been previously described for this sample.<sup>24</sup> Briefly, the sample consisted of individuals with a current condition of CP and individuals who never had CP or any of the following conditions that were available in the 2016 NSCH: epilepsy, blood disorders, genetic disorders, cystic fibrosis, Down syndrome, developmental delay, intellectual disability, autism, or severe difficultly walking. Several variables used in the current

investigation were not administered to children 5 years of age and younger. Therefore, the final sample was restricted to children aged 6 to 17 years old. Approval from an institutional review board was not necessary because the data used in this investigation are publicly available and deidentified.

Criteria commonly used to classify specific characteristics of CP were not available in the 2016 NSCH (e.g. the Gross Motor Function Classification System). On the other hand, the respondent's subjective report of the child's condition severity was available for all participants, and included either 'mild' or 'moderate or severe'.

#### **Outcome measures**

Respondents answered the following prompt: 'Has a doctor or other healthcare provider ever told you that this child has [specified disorder]?' The child was considered to have a mental health disorder if the respondent reported a current condition of depression, anxiety problems, behavioral/conduct problems, or attention-deficit/hyperactivity disorder. The child was considered to not have a mental health disorder if the respondent reported 'no' to the condition or the child had the condition, but not at the time of the survey. Multimorbidity was defined as having two or more individual mental health disorders.

#### **Predictor variables**

A dichotomous variable (yes/no) for participation in activities was determined if the child participated in one or more extracurricular organized activities or lessons after school or on the weekends in the past 12 months. Activities included sports teams or lessons, clubs or organizations, and any other organized activities or lessons.

A dichotomous variable (yes/no) for difficulty with friendships was determined by the following prompt: 'Compared to other children his or her age, how much difficulty does this child have making or keeping friends?' The child was defined as having difficulty with friendships if the respondent answered 'a little difficulty/a lot of difficulty'. The child was defined as not having difficulty with friendships if the respondent answered 'no difficulty'.

A dichotomous variable (yes/no) for bully victimization was determined by how well the following phrase described the child: 'This child is bullied, picked on, or excluded by other children'. The child was defined as experiencing bully victimization if the respondent answered 'definitely true' or 'somewhat true'. The child was defined as not experiencing bully victimization if the respondent answered 'not true'.

#### Covariates

Chronic pain is a problem in individuals with CP and is associated with lower quality of life,<sup>10</sup> behavioral and emotional problems,<sup>8,9</sup> and other mental health disorders, such as anxiety and depression.<sup>8</sup> Therefore, chronic pain was included as a covariate. A dichotomous variable for chronic pain (yes/no) was determined if the child had 'FREQUENT or CHRONIC difficulty' with 'repeated or chronic physical pain, including headaches or other back or body pain', as previously described. Other covariates included age, sex, ethnic group, and household poverty status as a ratio of the federal poverty line.

### Statistical analysis

To account for the multistage and complex survey design of the 2016 NSCH, we used sampling strata and the primary sampling unit to obtain correct variance estimation, and weighted estimates to account for oversampling, survey non-response, and distribution of the target population. Statistical analysis was performed using SAS version 9.4 (SAS Institute, Cary, NC, USA) with statistical significance determined by *p*-values less than 0.05 and two-sided confidence intervals (CIs). Unweighted sample size and weighted estimates of all variables as percentages (95% CI) are presented. Group differences for all categorical variables were tested using  $\chi^2$  tests. Data regarding the severity of CP are not validated. Therefore, this variable is reported with descriptive characteristics to better characterize the CP sample, but has not been used to adjust the statistical analyses. We also examined the correlation between the three predictor variables for the total sample using the phi coefficient.

Weighted multivariable logistic regression analyses were performed with mental health disorders (yes/no) as the dependent variable and group (CP vs controls) as the primary exposure variable. Model 1 adjusted for age, sex, ethnic group, household poverty status, and chronic pain. Then, the predictor variables were added to the multivariable logistic model based on the significance of correlation among the three predictor variables for the total sample. If the three predictor variables were significantly correlated with one another, they were modeled individually in addition to the variables in model 1 to reduce collinearity. The interaction of group by predictor variables was examined for each outcome; if not significant, it was removed from the model. Goodness of fit for each model was examined using the C-statistic. Since multivariable logistic regression is event-driven, we followed the recommendation by Peduzzi et

al.<sup>26</sup> and only performed multivariable logistic regression analysis when the number of events per variable was greater than 10 for each outcome to limit bias in estimating regression coefficients.

In the event that the odds of a mental health disorder were not significantly increased in children with CP after adjusting for the variables in model 1, subsequent models were not performed because the association between CP and that mental health disorder was accounted for. Interpretations are made based on the structure of the logistic regression models; that is, the predictors are the aforementioned social factors and the response variable are the individual mental health disorders. Individuals who had missing data for all variables in the logistic regression models, which represented less than 5% for CP and controls,<sup>24</sup> were included in the analyses.

# RESULTS

The descriptive characteristics of participants are presented in Table I. Of the children with CP, just over half (56.6%) had moderate or severe disability. There was no statistical difference between groups for age, sex, or household poverty status. There was a statistical difference between groups for the distribution of ethnic group (p=0.005), and children with CP had higher prevalence of chronic pain compared to controls (p<0.001). Children with CP had lower prevalence of participation in activities (p<0.001) and higher prevalence of difficulty with friendships (p<0.001) and bully victimization (p<0.001) compared to controls. In the total sample, participation in activities was significantly correlated with difficulty with friendships (p=0.014, p<0.001) and bully victimization (p=0.10, p<0.001); difficulty with friendships was significantly correlated with bully victimization (p=0.001).

Children with CP had higher prevalence of depression (7.8%, 2.7%; p=0.017), anxiety (30.2%, 6.2%; p<0.001), behavior/conduct problems (27.3%, 4.9%; p<0.001), attention-deficit/hyperactivity disorder (19.5%, 7.1%; p=0.030), and multimorbidity (22.3%, 5.2%; p<0.001) compared to controls.

The adjusted odds of mental health disorders are presented in Table II. After adjusting for the variables in model 1, the odds of depression (odds ratio [OR] 1.4; 95% CI 0.6–3.8; C-statistic 0.72) and attention-deficit/hyperactivity disorder (OR 1.7; 95% CI 0.6–4.6; C-statistic 0.64) were no longer significantly increased in children with CP; however, the odds of anxiety (OR 4.4; 95% CI 2.1–9.0; C-statistic 0.65), behavior/conduct problems (OR 3.9; 95% CI 1.4–11.3; C-

statistic 0.66), and multimorbidity (OR 2.8; 95% CI 1.1–7.0; C-statistic 0.61) were significantly increased in children with CP.

There was no significant group by predictor variable interaction (all p>0.05). When participation in activities was added to model 1, the odds of anxiety (OR 4.0; 95% CI 1.9–8.5; C-statistic 0.66), behavior/conduct problems (OR 3.7; 95% CI 1.3–10.4; C-statistic 0.67), and multimorbidity (OR 2.6; 95% CI 1.1–6.4; C-statistic 0.63) remained significantly increased in children with CP. When difficulty with friendships was added to model 1, the odds of anxiety (OR 2.1; 95% CI 0.9–4.6; C-statistic 0.75), behavior/conduct problems (OR 1.7; 95% CI 0.5–5.7; C-statistic 0.80), and multimorbidity (OR 1.1; 95% CI 0.4–2.9; C-statistic 0.77) were no longer significantly increased (i.e. attenuated) in children with CP. When bully victimization was added to model 1, the odds of behavior/conduct problems (OR 2.5; 95% CI 0.7–8.4; C-statistic 0.75) and multimorbidity (OR 1.9; 95% CI 0.7–4.8; C-statistic 0.72) were attenuated, while the odds of anxiety (OR 3.4; 95% CI 1.7–6.9; C-statistic 0.72) remained significantly increased in children with CP.

### DISCUSSION

The principal finding of this investigation is that social factors were associated with a higher prevalence of mental health disorders in children with CP after adjusting for sociodemographic variables and chronic pain. Specifically, difficulty with friendships accounted for the association between CP and anxiety, CP and behavior/conduct problems, and CP and multimorbidity. Bully victimization accounted for the association between CP and behavior/conduct problems and multimorbidity. Moreover, interactions of group by social factors were not significant for any of the mental health disorders in the present study, suggesting that children with CP demonstrate a similar pattern of association between these social factors and mental health disorders as their peers without CP.

Our finding of lower participation in activities in children with CP is consistent with previous studies.<sup>11,12</sup> Many factors can lead to low participation for children with CP. Physical factors (e.g. pain, low physical activity levels), developmental comorbidities (e.g. communication impairments), and environmental factors can independently or synergistically affect social participation. As potential evidence against our hypothesis, when participation in activities was added to model 1, the increased odds of selected mental health disorders persisted in children with CP (Table II). However, there is limited evidence to suggest that participation in and of

itself may not affect peer relations in this population because participation is not synonymous with inclusion. For example, there is evidence that parents' efforts to facilitate social activities for children with CP are not associated with quantity and quality of friendships.<sup>27</sup>

We found that 75% of respondents reporting on a child with CP reported that the child had difficulty with friendships compared to 17% of controls. Evidence suggests that children with CP value social and emotional networks,<sup>14–16</sup> especially friendships.<sup>15</sup> However, these children also report lower social support from friends compared to children without CP,<sup>13</sup> which is consistent with a previous study of children born preterm having fewer friends.<sup>28</sup> In the current study, when difficulty with friendships was added to model 1, the increased odds of selected mental health disorders were attenuated in children with CP (Table II). These findings suggest an association between difficulty with friendships and mental health disorders in children with CP. This is consistent with a longitudinal study that found friendships mitigated the risk of developing mental health disorders in adolescents without CP.<sup>17</sup> Therefore, it is important to identify the factors contributing to the gap between the desire and need for friendships<sup>14–16</sup> and the perceived fulfillment of friendships<sup>13</sup> in children with CP.

The importance of friendships on lessening the burden of mental health disorders includes an additional provision on the protective effects on being bullied.<sup>18</sup> Children and adolescents who have higher perceived support of friends have a lower propensity to bully or be bullied.<sup>29</sup> In the current study, children with CP had a higher rate of bully victimization compared to controls, with a prevalence in over half (55%) of the CP group. Children with CP are at an additional risk for bully victimization because of the physical vulnerabilities and social stigma associated with disability.<sup>19-21</sup> In the general adolescent population, bully victimization has detrimental effects on mental health, suicidal ideation, self-esteem, and overall quality of life.<sup>22,23</sup> Further, the association between bully victimization and mental health problems has been shown to be bidirectional.<sup>30</sup> In the current study, when bully victimization was added to model 1, the increased odds of anxiety persisted, but the odds of behavior/conduct problems and multimorbidity were attenuated in children with CP (Table II). These data suggest that bully victimization accounts for the CP-behavior/conduct problems and CP-multimorbidity associations. The findings support the need to improve bullying policies and develop programs that promote resilience to bully victimization in this population to improve mental health profiles.

#### **Study limitations**

First, this is a cross-sectional and observational study; thus, we could not determine causality or directionality in association between exposures and outcomes, as well as unmeasured confounding variables. Other comorbidities (e.g. intellectual disability, communication impairments, epilepsy) are prevalent in the CP population and are associated with risk factors for mental health disorders. Second, the NSCH survey method uses parent/guardian proxy reporting instead of self-reporting. Further, it was not possible to verify conditions through diagnostic evaluation or review of medical records. Therefore, conditions are subject to bias and misreporting. Third, commonly used criteria to classify CP characteristics, such as the Gross Motor Function Classification System, are not available in the NSCH survey, as previously noted. Pinquart<sup>19</sup> conducted a systematic review of bullying in children and found that children with more visible chronic physical conditions were more likely to have reported bully victimization compared to children with less visible chronic physical conditions. Future studies incorporating the degree and/or visibility of motor impairment (e.g. the Gross Motor Function Classification System) as a factor for the development of mental health disorders may provide further insight. Fourth, there were large differences in sample size between groups. However, the prevalence rate of CP in the 2016 NSCH is similar to previous reports from the USA, and the comparison group is a nationally representative group of children without developmental disabilities. Therefore, statistical analyses incorporating 'group' as the primary exposure variable in this study is representative of the CP population in terms of prevalence and provides clinical insight about the deviation of outcomes measures (e.g. anxiety) from what is typical in a nondisabled population of children.

In conclusion, the concerning prevalence of mental health disorders in children with CP<sup>24</sup> can be accounted for by modifiable social factors. Social development is a complex, dynamic, and interactive construct that is affected by many factors associated with CP. Lower participation was not associated with mental health disorders in children with CP in the current study; however, participation is not synonymous with full inclusion. Friendships are important to master developmental tasks and for psychosocial functioning in adulthood. Further, friendships protect against the mentally harmful effects of bully victimization, which is also associated with adverse mental health and functioning. These data may suggest the need for social and emotional support of friendships, promoting inclusivity among peers, and mitigating bully victimization or increasing resilience to bullying in children with CP.

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	СР	Controls	
	<i>n</i> =111	<i>n</i> =29 909	
0	Percentage (95% CI)	Percentage (95% CI)	р
Descriptive characteristics			
CP severity			
Mild	43.4 (25.3–61.6)		
Moderate or severe	56.6 (38.4–74.8)		
Age			0.31
6–11y	40.4 (21.9–58.9)	50.2 (48.9–51.6)	
12–17y	59.6 (41.1–78.1)	49.8 (48.4–51.1)	
Male	62.8 (46.4–79.2)	49.3 (48.0–50.6)	0.11
Ethnic group			< 0.01
Non-Hispanic white	60.7 (41.7–79.7)	51.5 (50.1–52.8)	
Non-Hispanic black	27.1 (8.1–46.0)	12.5 (11.6–13.5)	
Hispanic	10.8 (0.0-22.0)	25.7 (24.2–27.2)	
Other	1.4 (0.2–2.7)	10.3 (9.6–11.0)	
Household poverty status			0.67
0–199%	50.6 (32.3-68.9)	43.7 (42.3–45.1)	
200–299%	9.7 (2.8–16.5)	15.0 (14.1–15.9)	
300–399%	13.2 (3.5–22.9)	11.0 (10.4–11.6)	
≥400%	26.6 (9.6-43.5)	30.3 (29.3–31.3)	
Chronic pain	39.0 (20.6–57.3)	7.6 (6.9–8.3)	< 0.01
Social risk factors			
Participation in activities	49.6 (30.6–68.5)	80.5 (79.2–81.7)	< 0.01
Difficulty with friendships	75.2 (60.5–89.9)	16.7 (15.8–17.7)	< 0.01
Bully victimization	55.0 (36.7–73.2)	19.3 (18.2–20.3)	< 0.01

Table I: Descriptive characteristics and social factors by group

Weighted data and analyses are presented. CP, cerebral palsy; CI, confidence interval.

		Participation in	Difficulty with	Bully
	Model 1 <sup>a</sup>	activities and	friendships and	victimization
$\overline{\mathbf{a}}$		model 1	model 1	and model 1
U	OR (95% CI)	OR (95% CI)	OR (95% CI)	OR (95% CI)
Depression				
CP (reference:controls)	1.4 (0.6–3.8)	Not performed	Not performed	Not performed
Anxiety				
CP (reference:controls)	4.4 (2.1–9.0)	4.0 (1.9–8.5)	2.1 (0.9–4.6)	3.4 (1.7–6.9)
Behavior/conduct problems				
CP (reference:controls)	3.9 (1.4–11.3)	3.7 (1.3–10.4)	1.7 (0.5–5.7)	2.5 (0.7-8.4)
ADHD				
CP (reference:controls)	1.7 (0.6–4.6)	Not performed	Not performed	Not performed
Multimorbidity				
CP (reference:controls)	2.8 (1.1–7.0)	2.6 (1.1-6.4)	1.1 (0.4–2.9)	1.9 (0.7–4.8)

Table II: Multivariable logistic regression models for mental health disorders in children with CP

<sup>a</sup>Model 1: adjusted for age, sex, ethnic group, household poverty status, and chronic pain. Weighted data and analyses are presented. CP, cerebral palsy; OR, odds ratio; CI, confidence interval; ADHD, attention-deficit/hyperactivity disorder.