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Sleep Sufficiency in Pediatric and Adolescent Tourette's Disorder: National Survey of Children's Health

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ABSTRACT: *Objective:* The present study compared sleep sufficiency in youth with current Tourette's disorder (TD), history of TD and matched case controls, and examined predictors of sufficient sleep using a large US population-based survey. *Method:* Participants were 673 caregivers of youth aged 6 to 17 years (298 with current TD, 122 with a history of TD with no endorsement of current diagnosis, and 254 matched case controls) from the 2007 and 2011–2012 versions of the National Survey of Children's Health. History and current TD status, current comorbidity (attention deficit/hyperactivity disorder, anxiety, and depression) and psychiatric medication status were assessed by yes/no items. Current TD severity was dichotomized into mild or moderate/severe symptoms. Sleep was assessed by parent-reported number of sufficient nights their child slept in the past week. *Results:* Univariate analysis of variance yielded significant group differences in nights of sufficient sleep ($F[2,369.70] = 71.53, p < .001$), with controls having 1.5 more nights per week relative to both TD groups ($p < .001$). With respect to predictors of sufficient sleep, the analysis of covariance yielded a significant age \times sex \times TD severity interaction ($F[1,15.84] = 4.28, p = .04$) such that older adolescent males with mild TD had significantly fewer nights of sufficient sleep than children ($p = .004$) and early adolescents ($p = .002$; $F[2,54.93] = 7.45, p = .001$). Early adolescent females with moderate/severe TD had fewer nights of sufficient sleep relative to males ($p = .008$). Comorbidity type and psychiatric medication status did not significantly predict sleep. *Conclusion:* Findings suggest that insufficient sleep in youth with TD persists independently of comorbidity or psychiatric medication status. Findings highlight the importance of clinical sleep monitoring in this population.

(*J Dev Behav Pediatr* 0:1–5, 2017) **Index terms:** Tourette's disorder, sleep, comorbidity, medication.

Tourette's disorder (TD), sometimes referred to as Tourette syndrome, is a neurological disorder characterized by multiple involuntary movements (motor tics) and 1 or more vocalizations (vocal tics) lasting for a period greater than 12 months. Although both the Diagnostic and Statistical Manual of Mental Disorders [Fourth Edition, Text Revision] and DSM-5 refer to the condition as Tourette disorder, the National Survey of Children's Health [NSCH] surveys used the former name, Tourette syndrome; to best conform with current usage, Tourette disorder will be used throughout this report.^{1,2}

TD is prevalent in children at an approximate rate of 0.6%³ with symptoms presenting more frequently in males than in females at a ratio of 3:1.⁴ The average age of onset ranges between 5 and 7 years, with tics following a waxing and waning course which peaks in severity during early adolescence and steadily declines in severity through early adulthood years in many individuals.⁵ TD can exact substantial physical, social, emotional, family, and academic burden,^{6,7} and is associated with high rates of psychiatric comorbidity, particularly attention-deficit/hyperactivity disorder and obsessive compulsive disorder.⁸ Furthermore, sleep disturbance, including difficulties with sleep initiation, maintenance, and efficiency has frequently been demonstrated in TD, with rates as high as 65% in children with TD without psychiatric comorbidity. Moreover, greater sleep problems have been shown in TD relative to healthy control samples,^{9,10} with significant associations found between tic severity and sleep disturbance, and tics occurring during sleep in some individuals.^{11,12} Despite this overlap, reasons for the link between poor sleep and TD are unclear, with co-occurring psychiatric conditions, psychiatric medication, and neural circuitry described as possible mechanisms.¹³

Therefore, the primary objective of this study was to compare sleep sufficiency in youth with current TD, history of TD with no endorsement of current diagnosis,

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and matched case controls using a large nationally representative sample.^{14,15} Given the extant literature suggesting that youth with current TD report greater sleep problems than controls, we hypothesized that youth with current TD would have less sufficient sleep relative to those with a history of TD and control youth. In addition, we assessed demographic (age and sex) and clinical (severity of TD, psychiatric comorbidity, and medication status) predictors of sufficient sleep in the current TD group.

METHOD

Sample Origin

Participants were drawn from the National Survey of Children's Health (NSCH) 2007¹⁶ and 2011–2012¹⁷ cycles. The NSCH is a United States national survey centered on children aged 0 to 17 years. Survey collection is scheduled every few years, with completion thus far in the years 2003, 2007, and 2011–2012, and periodic data collection is ongoing. The 2007 edition of the NSCH was the first national, population-based youth survey to feature questions on TD, and both the 2007¹⁴ and 2011–2012¹⁵ TD samples have been previously profiled. The NSCH survey was funded by the United States Department of Health and Human Services, the Health Resources and Services Administration, and the Maternal and Child Health Bureau and conducted by the Centers for Disease Control and Prevention's National Center for Health Statistics, with the objective being to measure the physical and emotional health of children in the nation regarding a range of factors including family and parental well-being, school and extra-curricular activities, neighborhood systems, and access to and utilization of health care services.^{18,19}

Data were collected through the State and Local Area Integrated Telephone Survey (SLAITS) program,²⁰ for which households with at least 1 child under the age of 18 years were randomly selected from a random-digit-dial sample of landline telephone numbers supplemented with cell phone numbers. A parent or guardian familiar with the child's health information was invited to participate. One child from each household was selected to be the subject of the interview (with random selection in households with 2 or more children). A total of 91,642 interview surveys were collected for the 2007 sample and 95,677 surveys for the 2011–2012 sample from 1 child in each household. Translated versions of interviews were also provided in Spanish, Mandarin, Cantonese, Vietnamese, and Korean for non-English speakers. Data from the 2 cycles of the survey (2007¹⁶ and 2011–2012¹⁷) represent cross-sectional, non-overlapping households,^{18,19} which were merged in this study and not used for longitudinal analysis.

This study used the following NSCH items to address the research questions. To assess sleep, one item queried: "During the past week, on how many nights did [your child] get enough sleep for a child [his/her] age?" This item allowed for parent-report ratings of 0 to 7 nights of suffi-

cient sleep. Items assessing the history of TD, current TD, and current TD severity were also used. Note that although both Diagnostic and Statistical Manual of Mental Disorders (DSM; Fourth Edition, Text Revision superscript new) and DSM-5(superscript: #1) refer to the condition as Tourette disorder, the NSCH surveys used the former name, Tourette syndrome. Items include "Has a doctor or other health care provider ever told you that [your child] has Tourette Syndrome?", "Does [your child] currently have Tourette Syndrome?", which was asked only to those endorsing lifetime TD, and "Would you describe [his/her] Tourette Syndrome as mild, moderate, or severe?", which was asked only to those endorsing current TD. This item structure was also used to assess comorbid disorders (e.g., anxiety, depression, attention-deficit/hyperactivity disorder [ADHD], etc.).

Case Control Matching Procedure

To select a nonaffected group comparable in size to the TD group, the SPSS Case Control Matching procedure was used. Control youth were selected from those youth whose caregiver endorsed no current or history of psychiatric problems (anxiety, depression, ADHD, emotional or behavioral problems, and autism spectrum), no current or history of medications for any of the above-described conditions, and no current or history of mental health service use. The criteria used for this procedure (with match tolerance) included: (1) age (± 1 year), (2) sex (exact match), (3) year of survey (exact match), (4) race/ethnicity (exact match), (5) total children in household (± 1), and (6) current insurance type (government-assisted or private; exact match). We elected to match on these variables as they may be correlated with the primary variables of interest. For example, is it known that with increasing age (e.g., adolescence), there are naturally occurring biological and behavioral changes which are associated with decreases in sufficient sleep.²¹ In addition, sociodemographic/socioeconomic factors including race (i.e., African-American) and insurance type (i.e., lack of private insurance) are associated with increased sleep problems.²² Priority was given to exact matches, and they were case order randomized when selecting from more than 1 match. We also used the SPSS Case Control Matching procedure a second time, excluding the initially selected matched youth and re-ran analyses to ensure that our results were not simply due to a particular subsample of nonaffected youth. As analyses did not differ, we kept the first group selected through the matching procedure as our control sample.

Analytic Plan

All analyses were conducted using IBM SPSS Statistics 23 statistical software. After case control matching, descriptive statistics were examined to compare groups on demographic variables. A univariate analysis of variance, with Bonferroni-corrected post hoc pairwise comparisons compared youth with TD, a history of TD, and controls on nights of sufficient sleep in the past week.

Then, in the current TD group, demographic (age and sex) and clinical (current TD severity, psychiatric comorbidity, and psychiatric medication status) predictors of sufficient sleep were examined using the analysis of covariance. TD severity was defined as a dichotomous variable such that caregivers who indicated that their child's current TD was "moderate" or "severe" were ranked as 1, and those who indicated that their child's current TD was "mild" were ranked as 0. Psychiatric comorbidity was defined as a categorical variable, with youth either rated by their caregiver as currently being diagnosed with internalizing (depression and/or anxiety), externalizing (ADHD) or internalizing/externalizing (both ADHD and depression and/or anxiety), or having no comorbid diagnoses (reference group). Psychiatric medication status was rated as dichotomous (yes/no) and derived from the survey question "During the past 12 months, has [your child] taken any medication because of difficulties with [his/her] emotions, concentration, or behavior?" Nonsignificant predictors were removed, and interactions were tested sequentially based on the significance of individual predictor terms. Bonferroni correction was used to control for type I error.

RESULTS

Participant Characteristics

The final sample consisted of 673 youth (298 with current Tourette disorder [TD], 122 with a history of TD, and 254 matched case controls). Groups did not differ with regard to mean youth age ($F[2,51.54] = 2.66, p = .07$), sex ($\chi^2 = .001, p = .99$), primary language spoken at home ($\chi^2 = 1.39, p = .50$), or insurance coverage ($\chi^2 = 1.29, p = .86$). About ethnic/racial minority status, a greater proportion of youth with a history of TD were minorities (36%) relative to both youth with current TD (23%) and control youth (21%; $\chi^2 = 10.39, p = .006$). In regard to medication status, a greater proportion of youth with current TD (67%) and a history of TD (49%) answered "yes" to the question "Does [the child] currently need or use medicine prescribed by a doctor, other than vitamins?"

than control youth (17%; $\chi^2 = 133.68, p < .001$). See Table 1 for demographic information by group.

Aim 1: Compare Sleep Sufficiency in Youth with Current Tourette's Disorder, History of Tourette's Disorder with No Current Symptoms, and Matched Case Controls

A univariate analysis of variance indicated significant group differences in nights of sufficient sleep in the past week ($F[2,369.70] = 71.53, p < .001$). Post hoc pairwise comparisons with Bonferroni correction revealed that on average, the control group reported 1.5 more nights of sufficient sleep per week than either the current TD (mean difference = 1.51, SE = 0.14, $p < .001$) or history of TD (mean difference = 1.59, SE = 0.18, $p < .001$) groups, with no significant differences between youth with current TD and history of TD (Table 1).

Aim 2: Examine Demographic and Clinical Predictors of Sufficient Sleep in Tourette's Disorder

Analysis of covariance yielded a significant age \times sex \times TD severity interaction in predicting nights of sufficient sleep in the past week ($F[1,15.84] = 4.28, p = .04$; Table 2). Follow-up contrasts were examined to interpret this interaction, with age split into the mean (9.34–15.68) and plus (15.69–17.99) and minus (4–9.33 years) 1 SD from the mean for ease of interpretation. For females, there were no significant age group differences in nights of sufficient sleep for either those with mild TD ($F[2,0.46] = 0.06, p = .94$) or moderate/severe TD ($F[2,15.78] = 2.14, p = .12$). Similarly, for males with moderate/severe TD, there were no significant age group differences in nights of sufficient sleep ($F[2,1.13] = 0.15, p = .86$). However, for males with mild TD, older adolescents had significantly fewer nights of sufficient sleep than both children (mean difference = $-1.51, SE = 0.46, p = .004$) and early adolescents (mean difference = $-1.28, SE = 0.36, p = .002$; $F[2,54.93] = 7.45, p = .001$). See Figure 1 for graphical representations of the age \times sex \times TD severity interaction.

Visual inspection of the data also appeared to indicate that females might have fewer nights of sufficient sleep

Table 1. Demographic and Clinical Characteristics by Group

| | Current TD (n = 298) | History of TD (n = 122) | Controls (n = 254) |
|--|-------------------------|----------------------------|-----------------------|
| Age, mean (SD) | 12.51 (3.17) | 13.21 (3.28) | 12.48 (2.93) |
| Sex, % male | 80.2 | 80.2 | 80.3 |
| Minority status, % minority | 23.5 | 35.5 | 20.5 |
| Primary language spoken at home, % non-English | 4.0 | 6.6 | 4.3 |
| Insurance coverage, % government-assisted or private | 35.4 | 40.4 | 37.0 |
| Current need or use of medicine prescribed by a doctor, other than vitamins, % yes | 66.0 | 48.8 | 17.3 |
| Parent-reported nights of sufficient child sleep in the past week, mean (SD) | 5.49 (1.97) | 5.41 (2.21) | 6.59 (1.10) |

TD, Tourette's disorder.

Table 2. Predictors of Sleep Disturbance in TD: Age \times Sex \times TD Severity

| Independent Variables | F | Partial η^2 | <i>p</i> |
|---------------------------------------|------|------------------|----------|
| Age | 3.65 | .01 | .06 |
| Sex | 0.30 | .001 | .59 |
| TD severity | 0.08 | <.001 | .78 |
| Age \times sex | 0.01 | <.001 | .91 |
| Age \times TD severity | 0.15 | .001 | .70 |
| Sex \times TD severity | 3.18 | .01 | .08 |
| Age \times sex \times TD severity | 4.28 | .015 | .04 |

TD, Tourette's disorder. For sex, male sex serves as the reference group. For TD severity, mild serves as the reference group.

than males. Therefore, we also examined contrasts specifically comparing sex. These revealed that for early adolescents with moderate/severe TD, on average females had 4.23 nights of sufficient sleep as compared to males who had 5.82 nights of sufficient sleep in the past week (mean difference = 1.60, SE = 0.60, $p = .008$).

Comorbidity type (none, externalizing, internalizing, both internalizing and externalizing) and medication status did not significantly predict sleep in youth with current TD and therefore were excluded from the final presented model (Table 2).

DISCUSSION

This study is the first to assess sleep in a US population-based sample of youth with TD. Consistent with hypotheses, findings showed that parents of control youth endorsed 1.5 more nights of sufficient sleep in the past week relative to children with current TD or a history of TD. This is consistent with previous research showing greater sleep disturbance in those with TD relative to healthy controls.^{10,13,23} One benefit of the original survey is the inclusion of a question regarding the lifetime history of TD, which allowed for the approximation of youth with a history of TD but no endorsement of current TD diagnosis. No significant differences in nights of sufficient sleep were found

between youth with current TD or a history of TD; both groups reported fewer nights of sufficient sleep than controls. The lack of a difference between these groups may suggest that the sleep disturbance in those with TD goes above and beyond direct physical interference from tic symptoms and may be indicative of trait-level sleep disturbance that may not subside along with tic symptoms.

We also sought to evaluate potential demographic (age and sex) and clinical (severity of TD, co-occurring attention-deficit/hyperactivity disorder and internalizing symptoms, and medication status) predictors of sufficient nights of sleep in those with current TD. Results showed a significant age \times sex \times TD severity interaction, whereby among males with mild TD, older adolescents had significantly fewer nights of sufficient sleep relative to younger children and early adolescents. This is consistent with research demonstrating declines in TD severity and increases in sleep disturbance with increasing age in clinical samples.²⁴ Furthermore, among early adolescents with moderate/severe TD, females had fewer nights of sufficient sleep relative to males. Although research on the influence of sex on tic symptoms and impairment is limited in children, research in adults suggests a more severe symptom course and greater impairment in females relative to males^{25,26}; additional research might elucidate whether this impairment for females also extends to sleep. Neither comorbidity nor medication status predicted sufficient nights of sleep in youth with current TD in the present sample, suggesting that sufficient sleep in TD is not solely accounted for by co-occurring psychiatric symptoms and medication side effects. Although previous studies have shown that youths with TD and comorbid ADHD reported greater sleep disturbance relative to those with TD without psychiatric comorbidity^{9,12} and that sleep problems in children with tics are associated with internalizing symptoms,¹⁰ previous findings have also demonstrated that youth with TD without comorbid conditions demonstrate sleep deficits.⁹ Previous research studies also support the presence of sleep difficulties in TD regardless of psychiatric medication use.⁹ Future work should further examine

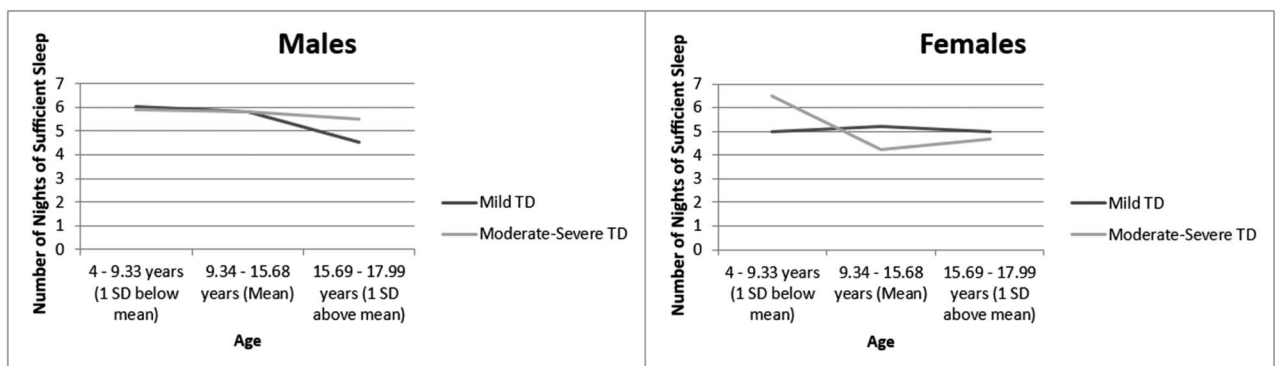


Figure 1. Number of nights of sufficient sleep: age \times sex \times TD severity. TD, Tourette's disorder.

the impact of age, sex, and TD severity on sleep deficits in this population.

Despite the sizeable group samples and nationwide representation, this study has limitations. First, because of the survey methodology used, we lack confirmation of TD diagnosis by a health professional. Also, TD severity was measured unidimensionally in this survey; however, tic severity encompasses multiple dimensions (e.g., tic number, frequency, intensity, complexity, and interference).²⁷ Moreover, the survey did not include a question on TD severity for those endorsing a history of TD, limiting the scope of the predictor analysis. In addition, the ratings are based on parent report as child perspective on sleep and tic symptoms was not assessed. Furthermore, parent ratings of sleep were retrospective and may be limited by recall bias. Finally, because of the cross-sectional nature of this work, we cannot address directionality in the relationship between sleep and tic symptom severity. Nevertheless, this study highlights the presence of insufficient sleep in youth with TD above and beyond the effects of comorbidity or medication use, and that youth with a history of TD may also experience insufficient sleep. This research suggests the importance of clinical monitoring for sleep disturbance in youth with current TD or history of TD.

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