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# Research in Autism Spectrum Disorders

journal homepage: [www.elsevier.com/locate/rasd](http://www.elsevier.com/locate/rasd)

## Characteristics of sleep in children with autism spectrum disorders from the Simons Simplex Collection

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### ARTICLE INFO

Number of reviews completed is 2

#### Keywords:

ASD  
Sleep  
Simons Simplex Collection  
Gastrointestinal disturbance

### ABSTRACT

**Background:** An estimated 40–80% of children with ASD have sleep problems, including bedtime behavior problems, difficulty falling asleep and staying asleep, decreased sleep time, and daytime sleepiness. This study aimed to examine the relationship between sleep problems and characteristics of children with ASD in a large, nationwide sample.

**Methods:** This secondary analysis of children 4- to 18-years explored 11 sleep problems using the Simons Simplex Collection Sleep Interview (SSCSI). The SSSCI includes nighttime problems, daytime problems, and sleep duration problems subscales. Chi square, Kruskal-Wallis and Mann-Whitney *U* tests were performed to detect differences between age groups, sexes, and sleep problem groups (none/minimal, mild, moderate/severe). Odds ratios for variables associated with sleep problems were assessed using baseline and adjacent category logistic regression. Two-way interaction effects were included in regression models, and stratified analyses were performed for age groups.

**Results:** Approximately 41% of children were categorized as having mild or moderate/severe sleep problems. The most commonly reported SSSCI items were in the nighttime problems subscale; difficulty falling asleep was the most frequently reported item. Mean sleep duration was approximately 9 h, although more than one-quarter slept less than the hours recommended for their age. Increased odds of sleep problems were most frequently associated with gastrointestinal distress (GID) and non-verbal IQ (NVIQ), followed by male sex and age. ADOS severity score is likely not associated with sleep problems in this sample.

**Conclusions:** This study advances our understanding of sleep in ASD by showing that GID, NVIQ, sex, and age increased the odds of sleep problems in children with ASD. These results reinforce that healthcare professionals should screen for sleep problems in children with ASD and suggest future lines of inquiry.

### 1. Autism spectrum disorder

Autism spectrum disorder (ASD) is a pervasive neurodevelopmental condition characterized by persistent difficulties in communication and social interaction, and restricted, repetitive behaviors, interests and activities. ASD is referred to as a “spectrum” because impairments may range from mild to severe, and may also include intellectual disability ([American Psychiatric Association, 2013](#)). The Autism and Developmental Disabilities Monitoring Network estimated that, in 2012, ASD affected 1 in 68 American children, and had a male-to-female ratio of 4.5:1 ([Centers for Disease Control and Prevention, 2016](#)).

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Received 9 October 2017; Received in revised form 2 June 2018; Accepted 4 June 2018

Available online 14 June 2018

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### 1.1. Sleep patterns in children with ASD

An estimated 40–80% of children with ASD have comorbid sleep problems (Reviewed in Veatch, Maxwell-Horn, & Malow, 2015; Cortesi, Giannotti, Ivanenko, & Johnson, 2010), which have been recognized by researchers for decades (Ornitz et al., 1969; Tanguay, Ornitz, Forsythe, & Ritvo, 1976). Studies have observed that, compared with typically developing [TD] peers, children with ASD have significantly longer sleep onset latency (time to fall asleep; Giannotti, Cortesi, Cerquiglini, Vagnoni, & Valente, 2011,  $N = 52$ ; Goldman et al., 2017,  $N = 41$ ; Lambert et al., 2016,  $N = 34$ ), more frequent awakenings after sleep onset (Allik, Larsson, & Smedje, 2008,  $N = 32$ ; Souders et al., 2009,  $N = 99$ ), shorter total sleep time (sleep beginning to end–awakenings after sleep onset; Elrod & Hood, 2015, meta-analysis  $N = 564$ ; Goodlin-Jones, Tang, Liu, & Anders, 2008,  $N = 194$ ; Miano et al., 2007,  $N = 62$ ), and poorer sleep efficiency (total sleep time/time in bed; all  $p < 0.05$ ; Elrod & Hood, 2015; Fletcher et al., 2016,  $N = 50$ ; Giannotti et al., 2011).

It has been suggested that intellectual disability (ID;  $IQ < 70$  with functional deficits; World Health Organization, 1996) may alter sleep patterns in children with ASD. Two recent studies have noted that ASD with ID was associated with sleep anxiety in children 2- to 5-years-old ( $p < 0.001$ ), bedtime resistance and night wakings in children 6- to 18-years-old ( $p < 0.05$ ; Mazurek & Petroski, 2015), and short sleep duration in children 4- to 18-years-old ( $p < 0.001$ ; Veatch et al., 2017). A meta-analysis of ten studies revealed that children with ASD and comorbid ID had significantly lower total sleep time than TD children (pooled difference of means =  $-45$  min; 95% CI:  $-62.6$  to  $-28.6$ ,  $p < 0.001$ ), while there was no significant difference in total sleep time between children with ASD and no ID compared to TD children ( $p = 0.316$ ).

Severity of ASD may also contribute to abnormal sleep patterns in children. In a population of 1224 children, those with parent-reported sleep problems (falling asleep and staying asleep) were observed to have higher ASD severity scores on the Autism Diagnostic Inventory-Revised social interaction, communication, and restricted, repetitive behavior subscales (Lord, Rutter, & Le Couteur, 1994); and on the Vineland Adaptive Behavior Scale (Sparrow, Balla, Cicchetti, Harrison, & Doll, 1984) communication, socialization, daily living skills, and motor skills subscales than children without sleep problems (Zachor & Ben-Itzhak, 2016;  $p < 0.02$ ). Using the Autism Diagnostic Observation Scale (ADOS; Hus, Gotham, & Lord, 2014; Lord et al., 1989) as an indicator of ASD severity, Veatch et al. (2017) observed that short sleep duration was significantly negatively correlated with ADOS social affect severity scores ( $\rho = -0.06$ ,  $p < 0.02$ ), though not with total ADOS severity score.

Two medical comorbidities that have been associated with sleep problems in children with ASD are gastrointestinal disturbance (GID) and seizures. Presence of epilepsy and epileptiform EEG abnormalities have both been reported as predictors of sleep problems (Giannotti et al., 2008;  $p < 0.01$ ). Aldinger, Lane, Veenstra-VanderWeele, and Levitt (2015) report that, among children with ASD and sleep problems (defined as parent report yes/no any sleep problems) in the Simons Simplex Collection (SSC), the odds of having GID (23% of sample) were almost two times higher than for children without sleep problems (OR = 1.8, 95% CI: 1.5–2.3), and the odds of having both GID and seizures (2.6% of sample) was twice as high as children without sleep problems (OR = 2.0, 95% CI: 1.3–3.1). In the Autism Genetic Resource Exchange, Aldinger et al. also report that children with sleep problems, seizures, and GID had a two-standard deviation lower Vineland Adaptive Behavior Scale composite score than children with none of the three comorbidities.

To date, most studies examining sleep problems in children with ASD have had small sample sizes and restricted age ranges, or look only at a single or select few sleep problems. The aim of this study is to examine the relationship between the presence of sleep problems and characteristics of children with ASD, such as age, sex, nonverbal IQ (NVIQ), ASD severity, and medical conditions in a large, nationwide sample of children 4- to 18-years-old. We hypothesize that the number of children with bedtime and nighttime sleep problems will decrease as children age; that children with GID and/or seizures will have more sleep problems; and that children with severe ASD and/or NVIQ will have more sleep problems.

## 2. Methods

### 2.1. Participants

#### 2.1.1. Simons Simplex Collection

The Simons Foundation Autism Research Initiative began in 2003 with the goal of improving the scientific understanding of ASD. One of their major projects was the Simons Simplex Collection [SSC], a sample of approximately 2750 simplex families (only one child with ASD) from 12 sites in the United States and Canada. Two studies have examined sleep patterns in children from the SSC. Veatch et al. (2017) examined short sleep duration in relation to intellectual and behavioral comorbidities of ASD, and Aldinger et al. (2015) studied risk for fourteen medical conditions, including presence of any sleep problem (yes/no), in children with ASD. The present study will examine ten sleep problems, including short sleep duration, and how these relate to demographic, medical, and behavioral characteristics.

Procedures of the parent study, inclusion and exclusion criteria, and characteristics of the SSC study sample have been described previously (Fischbach & Lord, 2010; Frazier, Georgiades, Bishop, & Hardan, 2014; Lord et al., 2012). Children, aged 4- to 18-years-old ( $N = 2745$ ,  $M = 9.0$ ,  $SD = 0.07$  years), were strictly phenotyped through parent interview, medical history, and multiple well-validated instruments. Children were given a clinical best estimate diagnosis of ASD based on observation, medical history and chart review, and Autism Diagnostic Observation Schedule [ADOS] and Autism Diagnostic Interview-Revised [ADI-R] scores. Diagnoses were based on the Diagnostic and Statistical Manual of Mental Disorders-IV-Text Revised [DSM-IV-TR] (Simons Foundation Autism Research Initiative, 2014).

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