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## Poor sleep affects daytime functioning in typically developing and autistic children not complaining of sleep problems: A questionnaire-based and polysomnographic study



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### ABSTRACT

Autism spectrum (AS) is a neurodevelopmental condition associated with poor sleep, which impairs daytime functioning. Most studies of sleep in autism have been based on subjective measures, notably parental reports. A few studies have used objective, laboratory polysomnography (PSG) measures, but often include confounding factors such as intellectual disability, sleep problems, other psychiatric illnesses, and medication. To address these limitations, we examined the relationship between sleep and behavior in prototypical AS of typical level of intelligence and non-autistic children not complaining of sleep problems. We examined sleep variables with The Children's Sleep Habit Questionnaire (CSHQ) and a daily sleep agenda, both filled out by parents, and by PSG. These subjective and objective measures both revealed that sleep latency was longer in AS than in non-autistic children. Furthermore, AS children also showed less slow-wave sleep (SWS: stages 3 + 4), fewer sleep spindles and fewer K-complexes than non-autistic children. REM sleep, including eye movement density, was similar between the two groups. The proportion of light sleep, (stage 1 non-REM sleep) was negatively correlated with IQ (Wechsler and Raven matrices) in both groups of participants. A large amount of SWS predicted low levels of internalizing behavior in both groups and typical social functioning as determined by ADOS in AS children. These results indicate that autistic children not complaining of sleep problems may nonetheless be affected by poor sleep, which in turn influences their daytime functioning.

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**Abbreviations:** ADI-R, autism diagnostic interview-revised; AS, autism spectrum; CSHQ, Children's Sleep Habit Questionnaire; HFA, high-functioning autism; IQ, intellectual quotient; PLMS, periodic limb movements during sleep; PSG, polysomnography; REM, rapid eye movement; SE, sleep efficiency; SWS, slow-wave sleep; TD, typically developing.

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## 1. Introduction

The autism spectrum (AS) is characterized by persistent alterations in social communication and social interactions across multiple contexts, as well as restricted and repetitive patterns of behavior, interests, and activities (American Psychiatric Association, 2013). Sleep problems are one of the most common comorbid conditions of AS (Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner, 2008; Patzold, Richdale, & Tonge, 1998; Richdale and Schreck, 2009). Between 50% and 80% of parents of children with AS report sleep problems, compared with between 10% and 25% of parents of non-autistic children (Allik, Larsson, & Smedje, 2006; Couturier et al., 2005; Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen, 2008; Malow et al., 2006; Polimeni, Richdale, & Francis, 2005; Richdale and Schreck, 2009; Souders et al., 2009). The most frequent sleep problems reported by parents of autistic children involve sleep onset, maintenance and duration (Cortesi et al., 2010; Reynolds & Malow, 2011; Richdale and Schreck, 2009), together with disturbances of circadian sleep (Glickman, 2010). Sleep problems in AS occur between early childhood and adolescence, and most parents (74%) using the CSHQ report such disturbances (Goldman, Richdale, Clemons, & Malow, 2012). Even in AS children of average intelligence, the prevalence and severity of sleep problems are higher than in non-autistic children (Couturier et al., 2005).

Most studies of sleep problems in autistic children are based on parental reports, which can be influenced by subjective biases, and few studies have used objective approaches to confirm the presence of sleep abnormalities in autism. Measures such as actigraphy are consistent with parental reports of sleep disturbances in autism, and confirm that sleep onset latency is longer and nocturnal awakenings are more frequent in autistic than in TD children (Goldman et al., 2009; Souders et al., 2009; Wiggs & Stores, 2004). However, actigraphy is not adapted to document long motionless periods of wakefulness, which can occur in some cases of insomnia (Sadeh & Acebo, 2002). In addition, this technique does not provide information about sleep stages, microarousals, rapid eye movements (REMs), and EEG phasic events such as sleep spindles and K-complexes. Polysomnography (PSG) is considered to be the gold standard for assessing the physiology and disturbances of sleep, but only a few PSG studies have compared the sleep autistic and non-autistic children (Bruni et al., 2007; Buckley et al., 2010; Dimedi et al., 1999; Elia et al., 2000; Goldman et al., 2009; Kulisek et al., 2008; Leu et al., 2011; Malow et al., 2006; Miano et al., 2007). These studies confirm the presence of longer sleep onset latency, lower total sleep time, lower REM sleep latency (Elia et al., 2000; Goldman et al., 2009; Miano et al., 2007) and less REM sleep (Malow et al., 2006) in autistic than in control children. By contrast, the percentage of SWS and the density of REMs are reported to be similar in non-autistic children and in children with autism (with or without intellectual disability), Down Syndrome or Fragile-X syndrome (Elia et al., 2000; Malow et al., 2006; Miano et al., 2007).

Most PSG studies of autistic children included individuals with intellectual disability (Buckley et al., 2010; Elia et al., 2000; Giannotti, Cortesi, Cerquiglini, Vagnoni, & Valente, 2011; Miano et al., 2007) or did not use an appropriate intelligence scale to assess the presence or absence of intellectual disability (Malow et al., 2006). Inclusion of individuals with intellectual disability may be an issue because sleep problems are common among individuals with low-functioning autism (Patzold et al., 1998; Richdale & Prior, 1995; Wiggs & Stores, 2004), and may involve psychophysiological mechanisms that are different from those involved in non-syndromic autism (Amiet et al., 2008; Moss & Howlin, 2009).

Three studies have investigated sleep in autism using PSG without influence from the confounding factors of medical comorbidities, intellectual disability and medication. Malow et al. (2006) defined AS children as “poor” or “good” sleepers according parental reports and found that poor sleepers showed a longer sleep onset latency and a lower sleep efficiency than good sleepers. Goldman et al. (2009) used PSG in a large sample of autistic children, including poor and good sleepers. They also found that AS children classified as poor sleepers had longer sleep latency than AS children classified as good sleepers, whereas other variables of sleep (total sleep time, sleep efficiency, wake time after sleep onset) did not differ between the two groups. Furthermore, there were no differences between AS and typically developing children classified as good sleepers. However, in these two studies, the classification of children as poor or good sleepers was based on a four-point parental concern checklist, not objective sleep measures. These two studies also included children with pervasive developmental disorders not otherwise specified (PDDNOS), which is a phenotype plagued with poor inter-experts reliability, and did not assess IQ. Finally, their calculation of sleep efficiency included the time in bed before sleep onset. The third study compared autistic adults without intellectual disability to group of neurotypical adults and reported a longer sleep latency, more nocturnal awakenings, a lower sleep efficiency, more stage 1, less SWS, lower amount of sleep spindles and lower densities of REMs in the clinical group (Limoges, Bolduc, Berthiaume, Mottron, & Godbout, 2013; Limoges, Mottron, Bolduc, Berthiaume, & Godbout, 2005).

A recent meta-analysis of the effect of sleep problems in typically-developed children demonstrated a strong association between sleep duration and cognition and internalizing/externalizing behavioral problems (Astill, Van der Heijden, Van Ijzendoorn, & Van Someren, 2012). Sleep problems in autism also affect daytime behavior and may be correlated with the manifestations of autism. Goldman et al. (2006) found that poor sleepers with AS tended to have more disruptive behaviors, such as hyperactivity and compulsive and ritualistic behavior, than good sleepers with AS. Malow et al. (2006) also point out that poor sleepers with AS present higher affective and attentional problems as well as anxious/depressed symptoms compared to good sleepers with AS and with TD children. Short sleep duration in autistic children was found to be associated with stereotypic behavior, social deficits, and overall high scores on autism scales (Allik et al., 2006; Malow et al., 2006; Patzold et al., 1998; Schreck, Mulick, & Smith, 2004; Taylor, Schreck, & Mulick, 2012). Sleep problems in autism are also associated with anxiety (Rzepecka, McKenzie, McClure, & Murphy, 2012), hyperactivity and oppositional behaviors (DeVincent, Gadow, Delosh, & Geller, 2007; Liu, Hubbard, Fabes, & Adam, 2006). Increases in behavioral difficulties related to

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