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Sleep problems in children with autism spectrum problems: a longitudinal population-based study

Børge Sivertsen
Division of Mental Health, Norwegian Institute of Public Health, Bergen, Norway Department of Clinical Psychology, University of Bergen, Bergen, Norway Division of Psychiatry, Helse Fonna HF, Haugesund, Norway

Maj-Britt Posserud
Centre for Child and Adolescent Mental Health, Uni Health, Norway Child and Adolescent Psychiatry, Haukeland University Hospital, Bergen, Norway

Christopher Gillberg
Gillberg Neuropsychiatry Centre, Sahlgrenska Academy, University of Gothenburg, Gothenburg, Sweden

Astri J Lundervold
Centre for Child and Adolescent Mental Health, Uni Health, Norway Department of Biological and Medical Psychology, University of Bergen, Bergen, Norway Kavli Research Centre for Aging and Dementia, Haraldsplass Deaconess Hospital, Bergen, Norway

Mari Hysing
Centre for Child and Adolescent Mental Health, Uni Health, Norway Department of Biological and Medical Psychology, University of Bergen, Bergen, Norway Betanien Hospital, Bergen, Norway

Abstract

This study examined the prevalence and chronicity of sleep problems in children who manifest problems believed to be typical of Autism Spectrum Disorders (ASD). Using data from a longitudinal total population study, symptoms of ASD, insomnia and potential explanatory factors were assessed at ages 7–9 and 11–13. Children were included in a group defined as having Autism Spectrum Problems (ASP) if they scored above a strict threshold on the Autism Spectrum Screening Questionnaire (ASSQ). Twenty-eight (0.8%) of 3700 children fulfilled the selected criteria for ASP at both waves, and the prevalence of chronic insomnia was more than ten times

Corresponding author:
Børge Sivertsen, Division of Mental Health, Norwegian Institute of Public Health, Bergen, Norway, Kalfarveien 31 5018 Bergen, Norway.
Email: borge.sivertsen@fhi.no

higher in these children compared to the controls. Children with ASP developed more sleep problems over time, with an incidence rate at wave 2 of 37.5% compared to 8.6% in the controls. The sleep problems were more persistent over time, with a remission rate of 8.3% compared to 52.4% in the controls. ASP was a strong predictor of sleep problems at wave 2 (OR = 12.44), and while emotional and behavioural problems explained a large proportion of this association, the effect of ASP on insomnia remained significant in the fully adjusted model (OR = 3.25). These findings call for increased awareness of sleep problems in children with ASP.

Keywords
autism spectrum disorders, emotional and behavioural problems, epidemiology, insomnia, sleep problems

Autism spectrum disorders (ASD) are a group of developmental disorders estimated to affect about 1% of children in the general population (Baird et al., 2006; Baron-Cohen et al., 2009; Honda et al., 2005). It is believed to exist on a distribution curve of autism spectrum problems (ASP) that exist in the general population, one for which there is no easily defined cut-off between subclinical and clinical presentations. ASP can have profound effects on both the child and his/her family. In addition to the core symptoms of social/communication deficits and a restricted repertoire of behaviours, children with ASD often experience sleep problems (Allik et al., 2006a; Richdale, 1999; Wiggs and Stores, 2004).

The majority of studies on sleep in children with ASD include clinical samples and use of cross-sectional design (Goodlin-Jones et al., 2008; Richdale and Prior, 1995; Wiggs and Stores, 2004). A population-based case–control study on ASD and sleep (Krakowiak et al., 2008) found a high rate of sleep onset problems and night waking in two- to five-year-old children with ASD compared with typically developing children. In a clinical follow-up study of school age children, higher rates of sleep problems were found in those with autism as compared with children with 'normal' development (Allik et al., 2008). Changes in sleep pattern over time were similar in both groups. There are also retrospective clinical studies indicating that sleep problems in children with ASD are persistent (Giannotti et al., 2006; Wiggs and Stores, 2004), but to our knowledge, there are no population-based studies on sleep in older children with ASD. There is also a paucity of longitudinal studies in the literature assessing the development of sleep problems over time in children with ASD. We know from studies of the adult general population that chronic sleep problems have a severe impact on both quality of life and physical and mental health (Taylor et al., 2005, 2007). As such, sleep problems in children/adolescents with ASD will most likely constitute an additional burden on both the children and their families, as they are already affected by the severe symptoms of ASD. In addition, there are studies indicating that insomnia in itself aggravates the autistic symptoms (Allik et al., 2006a; Schreck et al., 2004).

Emotional and behavioural problems are related to sleep problems in the general child population, and have also been reported to be associated with sleep problems in children with ASD (Richdale, 1999). In a previous study of children with Asperger's syndrome or high-functioning autism, those with insomnia were characterized by more co-existing emotional and behavioural symptoms than their peers, and more parent-reported autistic symptoms as measured by the Autism Spectrum Screening Questionnaire (ASSQ) (Allik et al., 2006a). Similarly, Patzold et al. (1998) found significant associations between sleep problems and problematic daytime behaviour in children with autism and Asperger's syndrome. However, the lack of longitudinal data and population-based studies has restricted the possibility of understanding the complex relationship between co-morbid emotional and behavioural problems and sleep difficulties in this group of children. To

target a potential need for increased sleep health care in children with ASD, sleep problems should be longitudinally studied in a total population setting. This is also important in order to obtain knowledge about risk factors and development of sleep problems over time, as well as for early prevention and detection.

Based on the above considerations, the aim of the current study was to assess sleep problems in children with ASP, here defined as scoring above a strict threshold on the ASSQ, when the children were 7 to 9 *and* 11 to 13 years old. Using data from a longitudinal population-based study, our specific aims were: (1) to assess the prevalence, remission rate and chronicity of sleep problems in children with ASP when compared with a large group of children without ASP; and (2) to identify risk indicators of sleep problems in children with ASP.

Methods

Participants

Data stem from the first and second waves of the Bergen Child Study (BCS), carried out in the autumn of 2002 and the spring of 2006, respectively. The BCS is a longitudinal total population study of children attending all the public and private schools in the city of Bergen, Norway. The protocol and the population of the BCS are described in detail elsewhere (Heiervang et al., 2007; Hysing et al., 2007). In short, in the first wave, the target population consisted of 9430 primary school children aged 7 to 9 years, for whom 7007 parents provided informed consent to participate. The second wave was performed four years later, and 5683 children, now aged 11 to 13 years, participated. A total of 4025 children participated with parental informed consent in both waves (70.8% of the originally consented population). Children with missing data on key variables used in this study were excluded from the analyses (n = 325), thus the total sample in the present study comprised 3700 children (47.3% boys).

Instruments

Autism spectrum disorder. The ASSQ (Ehlers et al., 1999) was used for all 3700 children at both times. It is a questionnaire designed to identify school age children who may need a more comprehensive evaluation because of suspected ASD. Identical versions of the ASSQ exist for parents and teachers. The instrument consists of 27 items for rating on a three-point scale; 'not true' (0), 'sometimes true' (1), and 'certainly true' (2). The items cover social interaction, verbal and non-verbal communication, restricted and repetitive behaviours, and motor problems including tics. The questionnaire was designed for completion by lay informants in school and home settings. The range of possible scores is 0 to 54. The ASSQ has been proven a reliable and valid instrument for screening individuals with ASD (across the range of intellectual abilities) in population and clinical settings (Ehlers and Gillberg, 1993; Ehlers et al., 1999; Posserud et al., 2009). Parent-teacher correlations have been shown to be $r = .66$ in clinical settings (Ehlers et al., 1999), and agreement measured by weighted kappa to be 0.40 in a general population setting (Posserud et al., 2006). Descriptive data from the ASSQ in the BCS have been published elsewhere (Posserud et al., 2008; Posserud et al., 2006, 2009). In the first wave of BCS a score of 17 or higher on the ASSQ was shown to yield both high sensitivity (0.91) and specificity (0.86) for a clinical diagnosis of ASD (Posserud et al., 2009). As the current study comprised no clinician-verified diagnosis of ASD, the term autistic spectrum problems is used throughout the current paper when referring to study's findings. ASP was operationally defined by an ASSQ score of 17 or higher *on both waves* by either a teacher or parent. Children with an ASSQ-score of 17 or more on one wave only were *not* defined as having ASP in the present study. Mental retardation was defined by the parent report, and coded as a dichotomous variable.

Sleep problems. Sleep problems were assessed by parental reports at both waves with one question encompassing difficulties with initiating and/or maintaining sleep, rated on a three-point Likert scale. A dichotomous variable was used for the purposes of the present study, in which responding either 'agree' or 'partly agree' was defined as having sleep problems. This operationalization has previously been applied in the BCS (Hysing et al., 2009; Sivertsen et al., 2009). Chronic sleep problems were defined as parent-reported sleep problems at both waves. No data on the severity of the sleep problems were available.

Emotional and behavioural problems. The Strengths and Difficulties Questionnaire (SDQ) was used in all 3700 children on both occasions. It is a behavioural screening questionnaire for children aged four to 16 years (Goodman, 1999, 2001). The 25 items describing positive and negative attributes of children can be allocated to five subscales with five items each: (1) emotional symptoms, (2) conduct problems, (3) hyperactivity-inattention problems, (4) peer relationship problems, and (5) pro-social behaviour. A total difficulty score is computed by combining the first four subscale scores. Each subscale is scored on a three-point scale; 'not true', 'somewhat true', and 'certainly true', with total subscale scores each ranging from 0 to 10, and total difficulties score from 0 to 40. The SDQ has been extensively validated in various countries (e.g. in population studies of children and adolescents in Nordic countries) (Heiervang et al., 2007; Muris et al., 2003; Smedje et al., 1999). The SDQ was completed by the parents in wave 1, whereas in wave 2 the SDQ was also provided by the children. The parent reports from wave 1 and the parent and child reports from wave 2 were included in the present study.

Demographic information

The level of parent education was reported in three categories (primary school, secondary school, and college/university), while household economy was rated as good, medium, or poor by the parents. Ethnicity was not reported as more than 90% of the Norwegian population is white (including immigrants) (Statistics Norway, 2008).

Statistical analysis

We first conducted Pearson chi-square tests and Kruskal–Wallis analysis of variance (ANOVA) with multiple comparisons to examine differences in demographics and clinical characteristics in children with and without ASP (ASSQ ≥ 17 on both waves). Chi-square without Yates correction was used to examine gender differences in children with ASP and sleep problems. We also examined children with an ASSQ score of 17 or higher on *one* of the waves (but not both). Non-parametric tests were chosen due to the non-normality of the data. Logistic regression analyses were used to further explore the association between ASP and sleep problems, and for these analyses, children with an ASSQ score of 17 or more on both waves were compared with children with ASSQ scores of less than 17 on both waves (children with an ASSQ score of 17 on *one* wave only were thus excluded). Both unadjusted (crude) analyses and separate analyses adjusting for potential explanatory variables were conducted. Results are presented as odds ratios (OR) with 95% confidence intervals (CI). Chi-square analyses were also conducted to examine the relationship between the severity of ASP symptoms and sleep problems. For analyses, the ASSQ sum scores (parent-reported) for each wave were spilt into four groups, with cut-offs based on the following percentiles: 2 = 25th percentile; 4 = 75th percentile, 10 = 95th percentile. Analyses were performed using SPSS for Mac 19, and the alpha level was set at a two-tailed 5%.

Ethics

The study was approved by the National Data Inspectorate and the Regional Committee for Medical Research Ethics in western Norway. Written informed consent was obtained from all parents included in this study. No payments were made for participation in the study.

Results

Sample characteristics

In the present sample, 28/3700 children (0.8%) were defined as having ASP with ASSQ scores ≥ 17 in both waves. Forty-three (1.2%) and 60 (1.6%) children fulfilled the criteria in the first or second wave only. There were significantly more boys in the ASP group (Table 1). Families of children with ASP had lower socioeconomic status with overall lower family income and lower parental education. Mental retardation was reported in 28.6% of the ASD sample, compared with only 0.1% among children with no ASP. Children with ASP also had more emotional and conduct problems, and a higher level of hyperactivity and inattention symptoms (see Table 1 for details).

Course of sleep problems in ASP

The prevalence of chronic sleep problems in children with ASP was 39.3% compared to 3.6% in the children with no ASP (Table 2). The prevalence rates of sleep problems reported only in the first or only in the second wave were 3.6% and 21.4% in the ASP group, compared to 4.0% and 7.9% in the non-ASP group. There was a significant association between sleep problems and severity of ASP symptoms, as illustrated in Figure 1, both at wave 1 ($\chi^2 = 179.14$, d.f. = 3, $p < .001$) and wave 2 ($\chi^2 = 230.3$, d.f. = 3, $p < .001$).

Children with ASD showed a chronic path of sleep problems with sleep problems at wave 1 being a stronger predictor for sleep problems at wave 2 (OR = 18.33, 95% CI: 1.86–179.90)

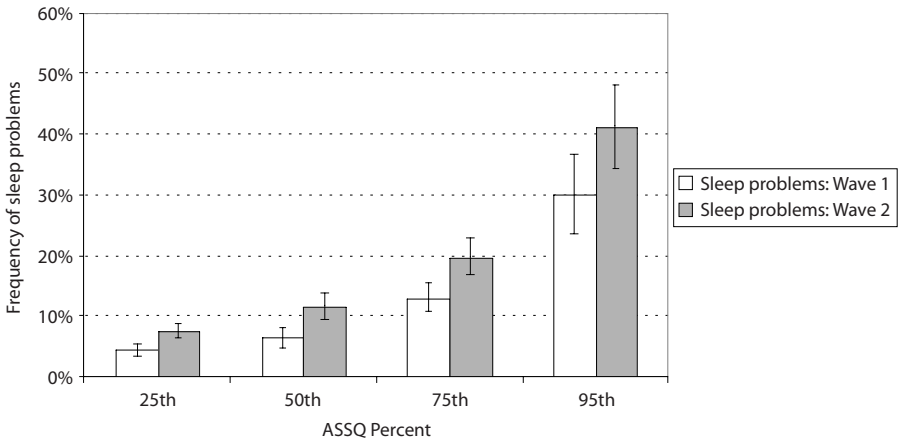

Figure 1. Frequency of sleep problems stratified by different percentiles of parent-reported Autism Spectrum Screening Questionnaire (ASSQ) sum score in both waves in the Bergen Child Study. Error bars represent 95% confidence intervals

Table 1. Demographic and clinical characteristics (wave 2) in children with Autism Spectrum Problems

Characteristics	Children with Autism Spectrum Problems (ASSQ (≥17)				
	No waves	1st wave	2nd wave	Both waves	P-value
N (%)	3569 (96.5)	43 (1.2)	60 (1.6)	28 (0.8)	
Boys, % (n)	46.5 (1660)	67.4 (29)	63.3 (38)	78.6 (22)	<.001
Mental retardation, % (n)	0.1 (4)	4.8 (2)	13.8 (8)	28.6 (8)	<.001
Family economy, % (n)					<.001
Good	71.3 (2436)	61.0 (25)	54.4 (31)	40.7 (11)	
Medium	26.7 (912)	26.8 (11|)	43.9 (25)	51.9 (14)	
Poor	2.0 (68)	12.2 (5)	1.8 (1)	7.4 (2)	
Education mother, % (n)					<.001
Primary	6.1 (207)	15.4 (6)	17.5 (10)	7.1 (2)	
Secondary	35.7 (1216)	35.9 (14)	50.9 (29)	50.0 (14)	
College/University	58.2 (1981)	48.7 (19)	31.6 (18)	42.9 (12)	
Education father, % (n)					.095
Primary	7.4 (248)	15.8 (6)	9.1 (5)	18.5 (5)	
Secondary	37.4 (1245)	39.5 (15)	45.5 (25)	33.3 (9)	
College/University	55.2 (1839)	44.7 (17)	45.5 (25)	48.1 (13)	
SDQ					
Emotional problems	1.05 (1.00–1.10)	2.26 (1.66–2.85)	2.83 (2.25–3.41)	4.04 (3.08–4.99)	<.001
Conduct problems	0.69 (0.65–0.72)	1.47 (1.07–1.86)	2.42 (1.93–2.90)	2.75 (2.01–3.49)	<.001
Hyperactivity	2.04 (1.98–2.11)	3.37 (2.68–4.06)	4.75 (4.09–5.41)	6.61 (5.63–7.58)	<.001

Table 2. Prevalence/incidence of and risk for sleep problems in children with Autism Spectrum Problems

	Children with Autism Spectrum Disorder	
	OR	95% CI
Sleep problems in 1st wave only		
Prevalence/incidence, %	3.6%	
Unadjusted odds-ratio (95% CI)	2.12	(0.27–16.70)
Adjusted odds-ratio (95% CI)*	1.62	(0.20–12.98)
Sleep problems in 2nd wave only		
Prevalence/incidence, %	21.4%	
Unadjusted odds-ratio (95% CI)	6.39	(2.30–17.72)
Adjusted odds-ratio (95% CI)*	5.99	(2.14–16.77)
Sleep problems in both waves		
Prevalence/incidence, %	39.3%	
Unadjusted odds-ratio (95% CI)	25.71	(10.73–61.63)
Adjusted odds-ratio (95% CI)*	16.43	(6.15–43.89)

* Odds ratios adjusted for sex, age, parental education and income

Table 3. Autism Spectrum Problems as a risk factor sleep problems at wave 2 adjusting for potential explanatory factors.*

	Sleep problems	
	OR	95% CI
Model A: Autism Spectrum Problems (ASSQ ≥17 at both waves)	12.44	(5.66–27.35)
Model B: A + Age, gender, parental education and income	9.62	(4.23–21.98)
Model C: B + Mental retardation	11.02	(4.69–25.91)
Model D: B + SDQ Emotional problems	4.71	(1.95–11.40)
Model E: B + SDQ Hyperactivity	4.25	(1.74–10.37)
Model F: B + SDQ Conduct problems	5.93	(2.45–14.35)
Model G: Fully adjusted (Model B+C+D+E+F)	3.25	(1.22–8.68)

* All explanatory factors were assessed at wave 2 of BCS

compared with the children with no ASP (OR = 9.86, 95% CI: 7.41–12.65). The remission rates was 8.3% in the ASP group, compared with 52.4% among children with no ASP ($p < .001$). As detailed in Table 2, ASP was a predictor for onset of sleep problems at wave 2, as well as for reporting chronic insomnia (both waves).

Co-morbidity of sleep problems in ASP

To examine potential explanatory factors accounting for the association between ASP and sleep problems, we conducted several analyses adjusting for different variables associated with ASP. In the unadjusted analyses, the OR for sleep problems in children with ASP was 12.44, and the risk remained at this level when adjusting for both demographics and mental retardation. When adjusting for emotional problems, the OR was reduced by 57%, and a similar pattern was found when adjusting for both hyperactivity and conduct problems. In the final model, adjusting for all of the listed explanatory factors, the effect of ASP on sleep problems was notably reduced, but remained an independent risk factor for sleep problems (see Table 3 for details).

Gender effects

Chronic sleep problems were more prevalent among boys than girls with ASP (50% (n = 11) versus 0%, $\chi^2 = 4.94$, d.f. = 1, $p = .026$). There was a similar gender effect in remission rates; all 11 boys with ASP and sleep problems at the first wave still reported sleep problems 3 years later, whereas the only girl with ASP and sleep problems at wave 1 no longer reported these problems at wave 2 ($\chi^2 = 12.00$, d.f. = 1, $p < .001$). No additional regression analyses were conducted separately for boys and girls due to the small sample of girls with ASP.

Discussion

In the present population-based longitudinal study, the development of sleep problems and emotional and behavioural problems in children with ASP was assessed when the children were between 7 and 9 years and 11 and 13 years. Children with ASP showed a higher rate of sleep problems at both time points compared with their peers. Their sleep problems were more chronic, showing a high stability over time, with a lower remission rate compared with sleep problems in children without ASP. Children with ASP also developed sleep problems over time at a higher rate than peers. The high rate of sleep problems could not be explained by sociodemographic factors or mental retardation, and while emotional and behavioural problems accounted for a large proportion of the association, ASP remained a significant risk factor for sleep problems in a fully adjusted model.

The current study is the first population-based study examining sleep problems in children with ASP using a longitudinal design, following a large group of children from middle childhood to early adolescence. Using a population-derived sample, our results demonstrate that ASP has an independent effect on sleep problems. These findings are in accordance with a population-based case–control study showing increased risk of sleep problems in preschool children (aged two to five) with ASD after controlling for various confounding factor (Krakowiak et al., 2008). In that study, the prevalence of sleep problems decreased from age two to five in both the ASD and the control group. In contrast, we found that the rate of sleep problems in the ASP group increased from age 7 to 9 to age 11 to 13 years. The current longitudinal population-based findings support the generally high prevalence rates of sleep problems (56–68%) in children with autism (Richdale, 1999), and extends on these findings by demonstrating the chronicity of the sleep problems in this group, and suggesting an increase in sleep problems over time. These results highlight the importance of assessment and treatment of sleeping problems as a standard and integrated part of the assessment and treatment of ASD. Although few studies have focused on behavioural interventions aimed at treating sleep difficulties in this population, some studies have shown that employing behavioural components may yield improvement in a variety of sleep problems (Richdale and Wiggs, 2005; Schreck, 2001). In terms of pharmacological interventions, melatonin has recently been shown to relieve insomnia symptoms effectively in children with ASD (Garstang and Wallis, 2006; Giannotti et al., 2006), and it has also been suggested that melatonin deficiency may be one of the main vulnerability factors leading to ASD (Melke et al., 2008). Diagnosing and treating insomnia in children with ASD is thus important both to relieve symptoms of autism, improve quality of life for the children and their families, and may even improve long-term outcome.

Looking at other risk factors for sleep problems in ASP children, the presence of hyperactivity symptoms as measured by the SDQ was a strong and independent risk factor for sleep problems in children with ASP. Children with ASD are at a much increased risk for having a

co-morbid attention deficit hyperactivity disorder (ADHD) or ADHD symptoms (Corbett and Constantine, 2006; Goldstein and Schwebach, 2004; Rommelse et al., 2010), and our study shows that this additionally increases the risk for sleep problems. This finding is in line with previous studies on children with ASD, which have also found a relationship between sleep problems and psychopathology, including ADHD (DeVincent et al., 2007). Although the direction of the association was impossible to examine in the present study, it is clear that examining and targeting ADHD symptoms in regards to sleep problems in this group is also important. In contrast to this, the presence of co-morbid mental retardation did not seem to cause increased sleep problems. This lack of impact of intellectual disability on sleep problems in children with ASD has also previously been reported (Hoshino et al., 1984; Patzold et al., 1998; Richdale and Prior, 1995).

Although not within the scope of the current paper, it should also be noted that the current study found a significant association of ASP with lower socioeconomic status. This is not a typical finding, as most papers generally find no or the opposite relationship with regards to family socioeconomic status in children with ASD (Allik et al., 2006a; Krakowiak et al., 2008)

In spite of very few girls in the ASP sample, we found that sleep problems were significantly less prevalent in girls than boys, and their sleep problems were also more transient. These findings are in contrast to previous studies on children with ASD in which no such gender differences in sleep problems have been demonstrated (Liu et al., 2006). However, this finding should be interpreted with caution, as the sample of girls with ASP and sleep problems was very small ($n = 6$). For this reason, it was not possible to conduct any statistical analyses beyond descriptive statistics and cross-tabulations.

There are several limitations of the present study. First, sleep problems were assessed by a joint variable (difficulties initiating or maintaining sleep), making it difficult to examine each construct separately, and we also had no measure of the severity of the sleep problems. However, most studies typically show that parental reports of a global sleep problem in children are usually confirmed by objective measures (Allik et al., 2006b; Wiggs and Stores, 2004), and there are studies of autistic children showing that subjective reports of sleep problems even tend to underestimate the problems compared with actigraphs (Goodlin-Jones et al., 2008; Oyane and Bjorvatn, 2005). Still, given the non-specific nature of this one included sleep question, our findings should be interpreted with some caution, and there is clearly a need for future epidemiological studies of children with ASD to include a better validated sleep questionnaire.

Another important limitation is the lack of ASD assessment. Although we relied on questionnaire-based data from the ASSQ as a proxy for the presence of autism symptomatology, we recognize that the ASSQ is a screening instrument and not a replacement for a clinician-verified ASD diagnosis. To minimize false positive 'autism spectrum' cases, we have used a very stringent definition, requiring a high ASSQ score in both waves 1 and 2. Scores of ≥ 17 on ASSQ have been shown to be associated with a very high risk for ASD (Ehlers et al., 1999; Posserud et al., 2009), and having such a high score on two occasions three years apart is likely to be an even stronger predictor of the presence of ASD. The prevalence of 0.8% does not indicate a severe overestimate of ASD cases. Furthermore, as ASD presently is being proposed as a spectrum problem, it is arguable whether a more categorical diagnostic approach would be more valid in this population-based study setting. The study shows a clear association between ASP and sleep problems, and the bias introduced by including false-positive children into the ASP group would tend to weaken this association. The true prevalence of sleep problems in clinically verified ASD is thus likely to be even higher. As such, the broad operationalization of ASP may actually be a strength of the study, as the results might be generalizable to a larger group. For example,

symptoms of ASD have been shown to be prevalent in a range of neurodevelopmental conditions (see Gillberg, 2010).

Conclusion

Children with autism spectrum problems have a high rate of sleep problems, also when adjusted for other mental health problems. Their sleep problems tend to be more chronic than in children without ASP. The findings call for increased awareness and development of treatment strategies of sleep problems in children with ASD.

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