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REVIEW

# Examining the Relationship Between Sleep Quality, Social Functioning, and Behavior Problems in Children with Autism Spectrum Disorder: A Systematic Review

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Abstract: Over forty percent of autistic children experience poor sleep quality, and social interaction difficulties are a core characteristic of autism. However, the relationship between sleep quality and social functioning and behavior remains poorly understood. This systematic review examined the evidence concerning the impact of sleep quality on the social functioning and behavior problems in autistic children and adolescents. It also identified key related factors and evaluated how this issue has been researched to date. Seven key journals were hand-searched and five databases were systematically searched, using keywords. Titles and abstracts of 4123 items were screened against eligibility criteria by two researchers. Relevant studies were retained if they were peer-reviewed empirical papers, published in English between 2000 and 2021. Then, the full text of 97 papers was screened and if they met the eligibility criteria, their reference lists were hand-searched. Forty-six studies were included in the final review. Data were systematically extracted and two authors critically appraised the strengths and weaknesses of studies using Critical Appraisal Skills Programme tools. Key themes were identified, because a meta-analysis was not possible due to the studies' heterogeneity. The review identified that sleep quality and social functioning are associated with one another and there is a small amount of evidence that a bidirectional causal relationship may exist. Evidence suggests that several nights of suboptimal sleep duration and a lack of deep continuous sleep negatively impact externalizing and internalizing behavior. Sleep quality is also reduced by anxiety and sensory sensitivity. However, longitudinal studies with larger samples are needed to establish causality. Future research needs to examine confounding factors and to develop consensus regarding best-practice processes for the objective measurement of sleep with autistic children. Additional research also needs to further examine the consequences of poor sleep quality on internalizing behavior, and the impact of socio-cultural practices.

**Keywords:** autism spectrum disorder, night waking, insomnia, behavior, anxiety, bedtime resistance

# Introduction

Autism is a neurodevelopmental disorder with a prevalence rate of 1 in every 68 children.<sup>1,2</sup> Autism is characterized by the core features of repetitive behaviors, restricted interests, affective and social interaction difficulties.<sup>3</sup> Social functioning has been defined as "the ability to navigate through the social world and form and maintain relationships that involve one's home, school, or workplace" (p2).<sup>4</sup> It can be challenging for autistic children to satisfy roles within an environment, relationships with family and friends, and social activities.<sup>4</sup> Autistic children differ in several ways from neuro-typical (NT) children regarding their social functioning.<sup>5</sup> They may be less likely to look towards social stimuli<sup>6</sup> or to initiate and share attention jointly with other people,<sup>7</sup> and individuals may exhibit less ability to recognize emotions, interpret vocal and visual cues,<sup>8</sup> and imitate behavior based on social cues.<sup>9</sup> Furthermore, autistic children use different strategies

to process facial information, emphasizing features and the mouth rather than focusing on the eyes. <sup>10,11</sup> Social functioning is also impacted by motivation, perception, anxiety, social skills, and emotional challenges. <sup>4,12</sup>

Sleep problems include dyssomnias that disturb the quality, amount, timing of sleep, including difficulty falling or staying asleep, excessive daytime sleepiness. <sup>13,14</sup> They also include parasomnias that involve physiologic events or behaviors that interrupt sleep, including arousal and partial arousal disorders or during the transition between sleep stages. <sup>14</sup> Sleep problems in children have been operationalized in research in terms of bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night-time waking, parasomnias, sleep-disordered breathing, and daytime sleepiness. <sup>15</sup>

Between 25% and 40% of NT children experience sleep disturbances and these have been associated with decreased social functioning in middle childhood and preadolescents<sup>16</sup> and in children aged 18–42 months.<sup>17</sup> A systematic review of studies that involved children aged 5–12 years found that shorter sleep duration was associated with internalizing and externalizing behavioral problems and having less complex cognitive skills and executive functioning ability.<sup>18</sup> Positive relationships between sleep quality/quantity, and better cognitive and/or behavioral outcomes have also been found for children aged 2–5 years.<sup>19</sup> Sleep disturbances occur in 40–83% of autistic children.<sup>20–23</sup> A recent meta-analysis examined studies that had measured sleep objectively using polysomnography (PSG) or actigraphy. This study found small but significant differences between autistic children and NT children regarding their sleep parameters. These differences included shorter total sleep time (TST), longer sleep onset latency (SOL), and reduced sleep efficiency (SE). However, there was no significant difference between the total sleep time (TST) in autistic children without intellectual disability (ID) and their NT peers.<sup>24</sup> Díaz-Roman et al<sup>25</sup> also conducted a meta-analysis of studies that included both subjective and objective sleep measures. They found that when sleep was subjectively measured, autistic children had significantly higher sleep anxiety, sleep onset delay, sleep onset latency, bedtime resistance, night awakenings, sleep-disordered breathing, parasomnias, and daytime sleepiness. The results determined using objective measures revealed autistic children had lower TST, a lower percentage of rapid eye movement (REM) sleep, longer SOL, lower SE, than the NT group.

In autistic children, poor sleep quality is negatively associated with physical and psychosocial wellbeing and quality of life,<sup>26</sup> and it is associated with behavior problems<sup>27</sup> and higher levels of hyperactivity, social withdrawal, and irritability.<sup>28</sup> Higher rates of externalizing and internalizing behavior are also associated with lower levels of social functioning in autistic children and adolescents<sup>29</sup> and NT children.<sup>30</sup> However, the impact of sleep quality on social functioning and behaviour problems, and the factors that moderate these relationships remain poorly understood. Further examination of this topic is warranted because sleep quality is a potentially modifiable predictor of social functioning and problematic behavior. Detailed knowledge about these relationships could inform caregivers, therapists, and clinicians who seek to improve the well-being of autistic children.

This systematic review aimed to increase understanding of the relationships between sleep quality and the social functioning and behavior problems of autistic children and adolescents. Its objectives were to examine the current evidence and identify the factors that underlie and mediate this relationship, and evaluate how this relationship has been examined to date.

#### **Methods**

# Search Strategy

The key search terms listed in Table 1 were identified using the patient, intervention, comparison, outcome search strategy tool.<sup>31</sup> Keywords and MeSH terms were used and adapted according to the requirements of the following databases and as they were searched: Medline, PubMed, Scopus, Psych Info, and the Web of Science. In addition, the following journals were hand-searched for articles that met the eligibility criteria which is summarized in Table 2: Sleep, Sleep Medicine, Behavioral Sleep Medicine, Nature and Science of Sleep Journal; Journal of Autism and Developmental Disorders, Autism, Autism and Research. Initial searches were limited to papers published between the years 2000–2021, and older articles were identified through hand searching the reference lists of articles that met the inclusion criteria. Papers were included if they were peer-reviewed empirical studies, published in English. They also needed to involve children

Table I Key Word Search Terms

Population		Outcomes/Phenomenon of Interest			
Autism	AND Children	AND Sleep Quality	AND Social Functioning and Behavior		
"autism" OR autistic' OR pervasive develop*' OR ASD OR "Asperger*" OR "autism spectrum disorder"	Adolescent* OR you* OR bab* OR infant* OR juvenile* OR kid* OR teen* OR toddler* OR child*	Nap* OR rest* OR sleep* OR "sleep duration" OR "night waking" OR "restless sleep" OR "poor sleep" OR "sleep disturbance" OR bedtime Or "wake time" OR "Sleep problem*"OR "sleep anxiety" OR "sleep latency" Or insomnia OR "sleep quality" OR "sleep efficiency"	"daytime disfunction" OR behavio* OR active* Or perform* OR action* OR "challenging behavio*" OR "social functioning" OR "communicat*" OR "gesture*" OR verbal* OR nonverbal OR non verbal		

Note: \*Search includes all the words beginning with the preceding letters.

Table 2 Eligibility Criteria

Inclusion	Exclusion
Peer-reviewed empirical studies published in English.	Papers that are unpublished, theoretical, review articles, case report articles.
Participants diagnosed with any type of autism, with or without ID and ADHD. The diagnosis can be parent reported.	
Participants must be children aged 0–17 years as part or all the population studied.	Studies involving adults aged 18 years and over.
Studies must focus on sleep quality – duration, night waking, falling asleep, or staying asleep.	Studies that do not focus on sleep quality.
Studies using any design and methodology.	
Studies must focus on social functioning or social functioning was included as an outcome.	Studies that do not focusing on social functioning and/or behavior problems.

diagnosed with any type of autism and to focus on sleep quality and social functioning. These searches identified 5270 items that were imported into an Endnote database. After duplicates were removed there were 4119 items.

# Study Selection

Firstly, two researchers worked independently (FB, RC), screening the title and abstract of each item against the eligibility criteria. This resulted in the retention of 97 papers, with 87% inter-rater reliability between researchers. Then, researchers (AM, SW) reviewed the papers on which FB and RC disagreed, and decided upon their eligibility. Following this, FB and RC independently screened the full-text of 97 papers, which resulted in an inter-rater reliability of 88%. SW and AM reviewed and discussed any papers on which there was disagreement and determined their eligibility. Finally, the reference lists of relevant papers were hand-searched for additional papers. The PRISMA flow diagram below (Figure 1) summarizes the results of the paper selection processes.

# Data Extraction and Quality Assessment

Two researchers (AM, SW) collaborated to extract details from the included studies, using a bespoke template table designed according to the review aims. Due to the heterogeneity of the studies, it was not possible to conduct a meta-analysis. Instead, the study contents were repeatedly examined and themes were identified.

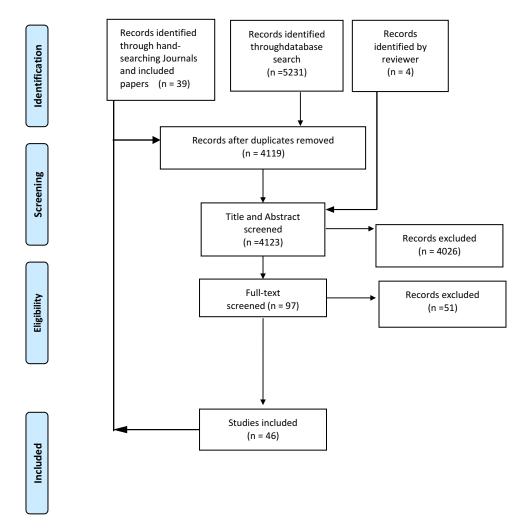


Figure I PRISMA Flow Diagram. Notes: Adapted from: Liberati A, Altman D, Tetzlaff J et al. The PRISMA statement for reporting systematic reviews and meta-analyses of studies that evaluate health care interventions: explanation and elaboration. Journal of Clinical Epidemiology. 2009;62(10)e1-e34. Creative Commons<sup>86</sup>.

The quality of the studies included in the review was assessed by researchers (RC, SG), who worked independently using the Critical Appraisal Skills Programme (CASP-uk.net) checklists, as recommended by World Health Organisation guidelines.<sup>32</sup> The CASP checklists are designed to assess the quality of research against criteria relevant to the design of the study under assessment. The CASP checklist for randomized controlled trials (RCTs) and the checklist for quantitative cohort studies include 12 points and the checklist for studies using qualitative methodology includes 10 points.

#### Results

Table 3 summarizes the content of the 46 studies that were included in the review and their key findings in relation to how sleep quality relates to social functioning and behavior in autistic children. The critical appraisal of the studies revealed that the methodological quality of these peer-reviewed studies ranged between moderate and high ratings on the CASP checklists. Due to the design of the studies, this review applied the RCT, Quantitative Cohort, and Qualitative checklists. The one qualitative study included in the review was rated 9 on the CASP Qualitative checklist; the four RCT studies were rated as between 10 and 12 using the CASP RCT checklist; the remaining 41 studies were rated as between 7 and 12 on the CASP Quantitative Cohort studies checklist. The inter-rater reliability between researchers' ratings during the quality appraisal was initially high at 86% and after discussions between researchers that also involved SW, 100% agreement was achieved.

 Table 3 Study Details (Abbreviations Key Below)

Study ID	Aims	Participant Details	ASD Diagnosis	Design and Methods	Measures	Key Results for ASD Participants	CASP
Aathira et al (2017) <sup>33</sup> India	Estimated prevalence and type of sleep problems associated with ASD severity, IQ, behavioral problems.	ASD (n=71), M (80.28%), aged 3-10, M = 5.3 SD = 1.8.	DSM IV	Cross-Sectional 2-yr study in teaching hospital. Parents were shown PSG prior to testing on for I night.	DP3, CBCL, CARS, CSHQ, cut-off 41, differentiated good sleepers and poor sleepers, PSG undertaken by N=48 (67.61%).	Poor sleepers had significantly more behavioral problems than good sleepers (p = 0.004).	П
Abel et al (2018) <sup>34</sup> USA	Examined the associations bet. sleep and challenging behaviors for average and night-to-night fluctuations.	ASD (n=39), M (84.62%), aged 2–10, M = 5.43, SD = 2.26.	SCQ	Cross-Sectional Behavioral treatment center. Parents instructed how to use the actigraph.	Daytime challenging behavior by the child's clinicians, Sleep diaries, actigraph.	Negative affect was associated with WASO ( $p < 0.001$ ) but not TST. Night waking associated with one additional negative affect per hour of waking ( $p < 0.001$ ).	10
Allik et al (2006) <sup>27</sup> Sweden	Compared ASD & NT on sleep/ wake behavior symptoms and problems.	ASD (n=32), M (87.5%), aged 8.5–12.8 M = 10.8	Independent clinician and ICD-10	Cross-Sectional Parents reported sleep patterns mail or clinic visits, teachers responded, via mail.	ASSQ, SDQ, Actigraph, Sleep diaries.	Children with insomnia showed higher hyperactivity (p= 0.04) and emotional symptoms rated by parents (p = 0.009) and teachers (p= 0.03), and lower pro-social behavior (p = 0.04).	10
Anders et al (2012) <sup>35</sup> USA	Examined relationships bet. sleep and daytime behaviors in comparison populations.	ASD (n=68), M (81%), aged 2-5.5 M = 47 Months SD = 10	ADOS & ADI-R	Longitudinal. Weeklong recordings completed enrolment, 3 and 6 months. After each recording week, parents reported sleep patterns.	Child Behavior Checklist CBCL, Actigraph, Sleep diaries, CSHQ.	Actigraph determined sleep problems not associated with behavior problems or daytime sleepiness. Parental reported of bedtime resistance associated with more daytime sleepiness (ps < 0.01) and behavior problems (ps < 0.01).	10
Chong et al (2021) <sup>36</sup> USA	Documented daytime electro- dermal activity (EDA) to explore their relations with sleep dysregulation/deficiency	ASD (n=13), M (80%), aged 3–9, M = 6.11 SD = 2.04 regulated sleep group (RegS) (n=6), dysregulated sleep group (DysS) (n=7)	SCQ, with a (>11) cut-off point	Cross-sectional Children recruited from 5 ABA treatment centers (N=5). Each child had received 20 hours per week ABA therapy.	VABS-II, Actigraphy, Sleep diaries.	DysS group had higher Social communication scores $(M=17.6, SD=4.28)$ than RegS $(M=22.5, SD=6.35)$ and were older $(p<0.01)$ . No definitive conclusions due to small sample.	9
Cohen et al (2017) <sup>37</sup> USA, Australia	Examined if sleep can predict adaptive functioning and ASD symptoms.	ASD (n=106), M (82.08%), aged 5–17 M = 14.77 SD = 3.11	DSM IV	Longitudinal 2 residential schools, caregivers instructed how to record the children's sleep quality.	VABS-II, Structured observations of sleep at 15–30 minute intervals.	"Unstable" sleepers showed greater significantly (p < 0.01) greater impairment in IQ scores and adaptive functioning. Socialization, communication compared to "Stable" sleepers.	12
Cohen et al (2018) <sup>38</sup> USA, Australia	Measured a predictive relationship bet. night-to-night sleep and day-to-day behavioral fluctuations.	ASD (n=67), Caucasian (88%), Hispanic (4.5%), Asian (4.5%), Native American (4.5%).	DSM IV	Longitudinal Participants from 2 residential schools. Sleep observed at 15–30 minute intervals. Clinician observed and recorded behavior during 1-on-1 daytime care.	VABS-II. Structured observations of sleep and behavior.	The accuracy of predicting behavior from prior sleep increased with the number of nights used to make the prediction. A significant ( $p < 0.05$ ) predictive relationship bet. priorsleep and daytime behavior in 81% of participants. Real-time fluctuations in sleep are important.	12

Table 3 (Continued).

Study ID	Aims	Participant Details	ASD Diagnosis	Design and Methods	Measures	Key Results for ASD Participants	CASP
Delahaye et al (2014) <sup>26</sup> USA	Investigate the relationship bet. HRQoL & sleep problems within the context of ASD characteristics.	ASD (n=86), M (83.72%), aged 4–12 M = 7.18	DSM IV & ADOS	Cross-Sectional.  Parents were recruited from an autism specialty clinic located at one of 3 centers. Parents notified firstly by phone.	CBCL & HRQoL, CSHQ, sleep- disordered breathing, daytime sleepiness	Shorter sleep duration ( $r = -0.36$ , p < 0.001), greater sleep anxiety ( $r = -0.29$ , p = 0.01) and parasomnias ( $r = -0.29$ , p < 0.01), had a consistent negative relationship with psychosocial aspects of HRQoL.	10
Fadini et al (2015) <sup>39</sup> Portugal	Investigated patterns of sleep and behavioral profiles in ASD children and adolescents.	ASD (n=45), M (78%), aged 4–18 M = 9.7 SD = 4.1	DSM V, ATA & CARS	Cross-sectional Questionnaires completed by parents and caregivers. And genetic tests completed.	CBCL, SDSC.	Positive correlation bet. thought problems and arousal disorders ( $r = 0.42$ , $p < 0.05$ ), excessive somnolence ( $r = 0.45$ , $p < 0.05$ ) and bet. Excessive somnolence and behavioral problems ( $r = 0.41$ , $p < 0.05$ ).	12
Fletcher et al (2017) <sup>40</sup> Australia, UK	Examined the association bet. individual changes in sleep profiles, anxiety, bedtime routines over 1-yr.	ASD (n=21) M (81%), aged 6–13 Baseline M = 106.67 SD = 26.83 months Follow-up M = 120.52 SD = 29.94	DSM IV	Longitudinal Actigraphs posted and documents emailed. Parents provided instruction. Movie vouchers given to child participants.	SWQ-P, CSHQ, Actigraph & Sleep diaries.	Those with increased frequency of maladaptive behaviors over time had increased SOL ( $r = 0.30$ , $p = 0.037$ ; Fisher's $r$ to z transformation, $Z = 0.48$ , $p = 0.63$ ). Improved sleep problems over 1 yr characterized by reduced sleep anxiety ( $r = 0.58$ , $p < 0.001$ ; Fisher's $r$ to z transformation, $Z = 0.17$ , $p = 0.34$ ).	11
Gunes et al (2019) <sup>41</sup> Turkey	Investigated the relationship bet. sleep difficulties and clinical symptoms of ASD.	ASD (n=112) M (72.3%), aged 2–18 M = 8.06 SD = 3.22	DSM V, CARS & dev. histories	Cross-sectional.  Assessments by a child dev. specialist and experienced psychologist. Sleep and social functioning parent-reported. If Denver II Test & WISC-R not possible used dev. histories, clinical symptoms observations, and average academic functioning.	AuBc, ABC-C & CARS, CSHQ. Denver II Test on children under 6 yrs. WISC-R full-scale on children over 6 yrs.	CARS total score correlated with Bedtimeresistance ( $r = 0.19$ , $p = 0.048$ ) and night wakings ( $r = 0.20$ , $p = 0.045$ ). No significant correlation bet. CSHQ scores and AuBC & AbBC total scores. Inattentiveness was significantly higher in children with CSHQ >56 ( $t(110) = -1.98 = p = 0.045$ ).	10
Goldman et al (2009) <sup>42</sup> USA	Determined if sleep concerns and objective measures are associated with daytime behavior.	ASD (n=42) M (90%), aged 4-10	ADOS	Cross-Sectional Recruited from Vanderbilt University Medical Center subspecialty clinics and the community	PCQ, CBCL, RBS-R, CSHQ, 2 consecutive nights of PSG & actigraphy monitoring video and EEG.	Poor sleepers had significantly ( $p < 0.05$ ) more inattention and hyperactivity than good sleepers. WASO correlated with hyperactivity ( $r = 0.48$ , $p = 0.04$ ) and sleep fragmentation correlated with RRB ( $r = 0.69$ , $p < 0.001$ ).	11
Goldman et al (2011) <sup>43</sup> USA	Identified variation in sleep- behavior across childhood- adolescence.	ASD (n=1784) M (84%), aged 3–18 M = 6.7 SD = 3.5	ADOS	Cross-Sectional.  Divided into good sleeper and poor sleeper and age cohorts <5 yrs, 5 to <7 yrs, 7 to <11 yrs, >11 yrs.	PCQ, CSHQ.	50% poor sleepers had problems with attention, social interaction, language, hyperactivity, sensory issues, anxiety, eating and self-stimulatory behaviors. 75% poor sleepers had problems with attention span, social interactions. I-unit increase on the parasomnia scale associated with 20% increase in the odds (95% CI) of problem regarding anxiety, sensory issues, aggression, hyperactivity, attention, mood swings, and SIB.	12

Henderson et al (2011) <sup>44</sup> USA	Examined the complexity of relations among routines general and bedtime, sleep quality and externalizing behaviors in ASD.	ASD (n=58) M (86.2%), aged 6-12 M = 9.0 SD = 2.09	CSBQ	Cross- Sectional. \$5 donated to an autism research foundation for participation.	CBCL, BRQ, CSHS, CSWS.	Consistency of bedtime routines was significantly positively correlated with sleep quality ( $r = 0.45$ , $p < 0.001$ ) and sleep quality was significantly negatively correlated with externalizing behavior ( $r = -0.39$ , $p < 0.01$ ).	10
Hoshino et al (1984) <sup>45</sup> Japan	To clarify the pathophysiological meaning of sleep disturbance.	ASD (n=75) M (86.6%), aged 3-15 years M=6 years 7 months.	Psychiatrist used WHO & Kanner criteria.	Cross-sectional	Psychiatrist Examination.	Day following sleep disturbance children took long naps (n = 26), loosing temper or low mood (n = 23), hyperactive (n = 9), lnactivity (n = 6), Isolationism (n = 3).	10
Johnson et al (2018) <sup>28</sup> USA	Assessed whether children with PS have greater daytime behavioral problems its impact on parental stress.	ASD (n=177) M (87.6%), aged 3-7 M = 4.7 SD = 1.14	DSM IV, ADOS, ADI- R	RCT -24-week Parents randomized to Parent Training or Parent Education Program. Questionnaires completed at study enrolment.	ABC, CSHQ. PSI.	Poor sleepers had significantly higher irritability (t(2.85), $p = 0.005$ ), hyperactivity/noncompliance (t(3.11), $p = 0.002$ ) than good sleepers.	10
Kang et al (2020) <sup>46</sup> China	Examined the relationship bet. sleep disturbances and emotional/ behavioral problems, and repetitive behavior	ASD (n=252), M (80.6%), aged 3-6 M = 5.13 SD = 0.097	DSM V & CARS.	Cross-Sectional Excluded if diagnosis of chronic illness, or mental disorder, or medications that could interfere with sleep.	SDQ, CSHQ, PPVT-C (completed by N=114 children, parents completed RBQ-2).	Highly significant correlations ( $p < 0.001$ ) bet. SDQ, CSHQ scores and sleep onset delay. Later had most ability to explain the variance (17.3%) in total SDQ score (adjusted R2 = 0.173). Moderate significant correlation ( $p < 0.01$ ) bet. sleep anxiety and SDQ.	11
Kelmanson et al (2020) <sup>47</sup> Russia	Evaluated sleep disturbances and their associations with emotional/ behavioral problems	ASD (n=18), All males aged 5 yrs	No Details provided	Cross-Sectional.  Parents requested to fill in questionnaire.	CBCL, CSHQ.	Emotional behavioral problems were poor predictors of sleep disturbance but the latter predicted probability of anxiety and affective problems.	9
Kirkpatrick et al (2019) <sup>48</sup> Ireland	Used a qualitative approach to explore parental perceptions of insomnia.	ASD (n=15) M (93.3%), aged 4-12 M = 8 SD = 2.17	Parent reported	Cross-Sectional. Study advertised with posters adverts in social media groups and websites and support services.	CSHQ, Focus groups (n=2)	Children (86%) experienced sleep anxiety that resulted in delayed bedtimes, night waking, and disruptive night behaviors. Poor sleep resulted in reduced daytime interactions (60%), aggressive outbursts (26%) and limited educational opportunities (87%).	9
Krakowiak et al (2008) <sup>49</sup> USA	Examined sleep characteristics using large sample comparing ASD, NT, and Children with Developmental Delay.	ASD (n=303), M (55.16%), aged 2–5 M = 3.6 SD = 0.08	ADI-R & ADOS	Case-Control Study  Data collected from California birth records, telephone interviews, clinic visit, and parent- administered questionnaires.	VABS, The CHARGE Sleep History parent- reported questionnaire, (similar to CSHQ).	Parents reported poor sleep affected child's daily functioning (21%) and family functioning (23%).  Sleep disturbances not associated with cognitive and/or adaptive delays.	9

(Continued)

Whelan et al

Table 3 (Continued).

Study ID	Aims	Participant Details	ASD Diagnosis	Design and Methods	Measures	Key Results for ASD Participants	CASP
Lambert et al (2016) <sup>50</sup> Canada	Assessed sleep and sleep-daytime functioning relationships in ASD children without subjective sleep Complaints	ASD (n=11), aged 6–13 M=10.27 SD=2.24.	ADI-R & ADOS	Cross-sectional.  Participants screened via a telephone interviews.  Participants resumed their normal daily activities the morning after PSG.	CBCL, CSHQ, sleep agendas, PSG (2 consecutive nights- 1st night for adaptation, data from 2nd night).	Proportion of SWS negatively correlated with reciprocal social interaction score ADOS ( $r = -0.807$ , $p = 0.003$ ) and positively correlated with repetitive behavior ( $r = 0.714$ , $p = 0.02$ ). Low amounts of SWS were associated with high levels of internalized behaviors ( $r = -0.41$ , $p = 0.046$ ).	10
MacDuffie et al (2020) <sup>51</sup> USA	Characterized sleep patterns and associations with brain dev. in children at risk/low risk of ASD and those who did/did not have ASD.	ASD (n=38), M (76%), aged 0-2, M=23.6 SD=3.9	ADOS	Longitudinal. Infants assessed at 6, 12, 24 months. MRI scans, parent-report measures, and measures of cog. and adaptive functioning.	ADOS, IBQ-R, VABS.	Sleep problems were not significantly related to social-affective symptoms severity ( $r_8$ = 0.26, $p$ = 0.15). Children with more sleep problems had higher trajectory of repetitive behavior over time ( $\chi^2(1)$ = 5.59, $p$ = 0.02).	Ш
Malhi et al (2019) <sup>52</sup> India	Examined association of sleep disturbances with daytime behavioral difficulties in ASD children.	ASD (n=60), M (85%), aged 4–12 M = 6.1 SD = 2.4	DSM V	Cross-sectional. Dev. behavioral, and sleep history taken. Parents asked typical time bed and wake times, total sleep duration at night, and the number of times child wakes at night, and total duration of night awakening.	DP 3, CSHQ.	Sleep dysfunction had a significant relationship with daytime reported behavior difficulties ( $r = 0.53$ , $p = 0.01$ ).	10
Malow et al (2012) <sup>53</sup> USA	Assessed dose–response, tolerability, safety, feasibility of Melatonin Supplement.	ASD (n=24), M (83.3%), aged 3–10 M=5.9 SD = 1.9/2.3	States children had an ASD clinical diagnosis details not provided	RCT- 14-week intervention.  Week1 (baseline), parents given 1-hour sleep ed.  During week 2 acclimation) children given inert liquid before bedtime. Children given melatonin (1mg) before bedtime for 14-weeks, increased to max. 9mg until satisfactory response.	CBCL, Sleep diaries and Actigraphy.	After treatment there were significant decreases $(p < 0.0001)$ in sleep latency, affective problems and social withdrawal. Significant decreases in ADHD $(p = 0.006)$ , Stereotyped behavior $(p = 0.008)$ and Difficult child $(p = 0.003)$ . Sleep duration significantly increased $(p < 0.0001)$ .	11
Manelis-Baram et al (2021) <sup>54</sup> Israel	Examined the longitudinal relationship bet. sleep disturbances and sensory sensitivities.	ASD (n=103) M (76%), aged 3–5 Time I M = 3.04 SD = 1.12, Time 2M = 4.47 SD = 1.19.	ADOS-2	Longitudinal.  Convenience sample at 2 separate time points.  Quantified changes in sleep disturbance and  Sensory Profile scores by subtracting Time 2  scores from Time 1 scores.	ADOS-2, CSHQ, SP	Longitudinal changes in sleep disturbances correlated significantly with sensory sensitivities ( $r = 0.42$ , $p < 0.001$ ) but not changes in ASD severity ( $r (57) = -0.04$ , $p = 0.75$ ).	10
May et al (2015) <sup>17</sup> Australia	Examined changes in sleep disturbances over 1 yr and relations bet. dev. change in sleep and behavioral problems.	ASD (n=46) M (52.17%), aged 7-12 M=9.84 SD=1.89	DSM IV	Longitudinal. Parents completed questionnaires at baseline and lyr later).	SRS, CSHQ.	Sleep quality scores from Time 1 and 2 respectively correlated significantly (p < 0.01) with anxiety ( $r = 0.585$ ; 0.701), social responsiveness ( $r = 0.581$ ; 0.455) and aggression ( $r = 0.281$ ; 0.261).	10

Mazurek et al (2019) <sup>55</sup> USA	Examined the chronicity and longitudinal relations of sleep problems and co-occurring symptoms.	ASD (n=437) M (82.61%), aged 2–10 M=5.07; SD=2.14	DSM IV & ADOS	Longitudinal. Participants evaluated by clinicians. Participants assessed in accordance with relevant measures at baseline then 3.8 years later.	CBCL, CSHQ.	Sleep problems significantly correlated (p < 0.01) with aggression ( $r = 0.431$ ), anxiety ( $r = 0.582$ ), somatic complaints ( $r = 0.435$ ), Sensory overresponsivity ( $r = -0.602$ ).	12
McLay et al (2021) <sup>56</sup> New Zealand, USA	Investigated the effects of function- based behavioral sleep interventions on ASD severity, behavior, parent relationship and sleep quality, depression, anxiety, stress.	ASD (n=41), aged 2–18 M=7; SD=3.4	Verified by psychologist, psychiatrist, or pediatrician.	RCT. These data were collected from child and adult participants who completed the initial clinical assessment and the prescribed treatment and questionnaires administered pre- and post-intervention.	CBCL, GARS. VABS-II PSQI.	Moderate sleep intervention effect for social interaction (ES = $-0.51$ , PSES = $74\%$ ). Small effect for emotional response (ES = $-0.35$ ; PSES = $61\%$ ), social communication (ES = $-0.25$ ; PSES = $66\%$ ). Improved internalizing (PSES = $73\%$ ) and externalizing behaviors (PSES = $69\%$ ).	10
Mutluer et al (2016) <sup>57</sup> Turkey	Investigated sleep problems to identify the risk/protective behavioral factors.	ASD (n=64), M (79.68%), aged 6-18 M = 11.66 SD = 3.8	DSM V	Cross-Sectional. Assessments completed by researchers.	ABC, PSQ, CARS, ABC, PSQ, CBCL.	Behavior problems correlated significantly with sleep problems ( $r = 0.597$ , $p < 0.05$ ) Social withdrawal and inappropriate speech variables were significantly correlated with sleep problems.	П
Naito et al (2019) <sup>58</sup> Japan	Investigated the time course of body movements at night and their relationship with social ability.	ASD (n=17), M (76.47%), aged 5–8 M=6.43	DSM V, ADOS	Pilot Study.  Accelerometer attached to the waist for 3 nights.	SRS, Vineland-11 MBS, Accelerometer:	Higher rate of body movement at 0.5 to 1 hour after onset of body stillness was associated with lower social ability ( $r = -0.507$ , $p = 0.036$ ) and maladaptive behavior ( $r = -0.529$ , $p = 0.028$ ).	10
Ng et al (2020) <sup>59</sup> USA	Assessed associations bet. sleep disturbances and externalizing/ internalizing problems, ADHD & social impairment.	ASD (n=28), ASD & ADHD (n=57), aged 7-17 M = 11.18 SD = 2.90	DSM V, ADOS-2	Retrospective Study. Informant inventories administered during child's evaluation. Participants had not been previously diagnosed with sleep apnea.	CBCL, PSQ. Psychiatric, Neuropsychological, Speech and Language evaluations.	In ASD and ADHD group sleepiness concerns correlated with externalizing problems (r = 0.39, p = 0.003). No associations bet. behavioral ratings and sleep in ASD group (rs < 0.36, ps > 0.061). Sleep factors contributed the variance in externalizing problems (17%), hyperactivity/ impulsivity (16%), in attention (15%).	12
Papadopoulos et al (2019) <sup>60</sup> Australia UK	Evaluated efficacy of a brief behavioral sleep intervention program.	ASD (n=61), ADHD- ASD (n=28) M (89%), Intervention, n=33 usual care aged 5–13 M=10.05 SD=1.85	Diagnosed by questioning parents.	RCT.  Measures administered baseline and 3 and 6 months. Intervention 2 face-to-face sleep consultations. 2 follow-up tel calls 2 weeks apart with trained clinician	Parent & teacher- reported SDQ, CSHQ, PedsQL, DASS, Sleep diary.	Difficult daytime behavior significantly correlated with sleep problems $r=0.42,p<0.01,$ night waking $r=0.49,p<0.01,$ and poorer sleep quality $r=0.45,p=<0.01.$	11
Patzold et al (1998) <sup>61</sup> Australia	Examined the specificity of sleep problems and their. Relationship with daytime behavior.	ASD (n=38) M (81%), aged 44–152 months SD=31.5	DSM-III criteria.	Cross-sectional.	14-day parental reported sleep diary. DBC, CBCL	Behavior problems correlated significantly with sleep problems ( $r = 0.42$ , $p < 0.01$ ), poorer sleep quality ( $r = 0.45$ , $p < 0.01$ ), night waking ( $r = 0.49$ , $p < 0.01$ ).	10

(Continued)

Whelan et al

# Table 3 (Continued).

Study ID	Aims	Participant Details	ASD Diagnosis	Design and Methods	Measures	Key Results for ASD Participants	CASP
Phung & Goldberg (2017) <sup>62</sup> USA	Examined the association bet. nocturnal sleep problems and daytime sleepiness in relation to quality of peer relationships.	ASD (n=19), M (84.2%), aged 5-13 M=16.88 SD=2.50	SCQ & ADOS-2	Cross- sectional.  Home visit, adolescents completed surveys and instructed re. actigraph sleep watch. 2nd visit materials collected and participants compensated.	NRI-RQV, Actigraphy 7 nights, SHS, Sleep diary.	Night-time and daytime sleep-wake problems were significantly associated with discordant relationships ( $r = 0.59$ , $p < 0.01$ ). Poor sleep associated with more perceived conflict with peers and daytime sleepiness.	9
Saré & Smith (2020) <sup>63</sup> USA	Examined relationship bet. sleep problems and autistic behavior.	ASD (n=29,276) M (80%), aged <18 M=8.6 SD=4.2	scQ	Cross- sectional.  Analyzed data from SPARK database	scq.	Significant association bet. repetitive behaviors and sleep problem ( $p < 0.001$ ) Slightly stronger association in females vs males.	7
Schreck et al (2004) <sup>64</sup> USA	Examined sleep problems and relations to the expression of ASD features.	ASD (n=55), aged 5–12 M=8.2 SD=2.1	GARS Autism Quotient ≥ 80)	Cross- sectional.	GARS, BEDS.	Shorter sleep duration predicted difficulties with social interactions ( $R^2=0.12,p<0.01$ ) and ASD diagnostic characteristics ( $R^2=0.11,p<0.02$ ). Communication problems were significantly related to periods of screaming during the night ( $R^2=0.18;p<0.01$ ).	II
Schroder et al (2019) <sup>65</sup> France, USA, Netherlands, UK, Israel	Examined effects of melatonin MedPRM on behavior and caregiver well=being outcomes in ASD children with insomnia.	ASD (n=125), M (75%) Placebo n=65 M n=47 (72.31%), aged 2–17 M=8.4; SD=4.24 n=60 MedPRM Aged 2–17 M=9; SD=4.08	DSM IV/V	RCT.  2-week single-blind placebo run. If impaired sleep participants randomly assigned to placebo/ drug group for 13 weeks as double-blind treatment.  Dose for efficacy 2mg, 5mg, 10mg.	SDQ, Sleep diaries.	After the 13 weeks treatment sleep improved in 41% children and there were significant improvements of SDQ mean behavior attributes ( $p = 0.077$ ). Significant improvement in externalizing ( $p = 0.021$ ) but not internalizing behavior ( $p = 0.770$ )	12
Segawa et al 1992 <sup>66</sup> Japan	Examined the effects of early disturbances in the S-W cycle on behavioral abnormalities.	ASD n=27	DSM II-R By Psychiatrists.	Longitudinal. Monthly or Bimonthly recording for 4 years. Parents educated how to encourage developing a 24-hour cycle.	Psychiatrist examination and parent sleep/wake diary.	In most children, stabilization of the sleep-waking cycle occurred before improved behavior but no significant correlation bet. age of improvement in sleep and behaviors.	9
Thenhausen et al (2017) <sup>67</sup> Germany	Investigated sleep problems in individuals with ASD and Asperger's Syndrome (AS).	ASD (n=15) M (86.7%), aged 10-17 M = 14.32 SD=3.03	Independent psychiatrist and psychologist	Pilot Study. Information sheet and questionnaires given by therapists to parents at home and returned personally or via mail.	SRS, SDSC.	The total sleep disturbance score correlated significantly with impairments in social communication ( $r = 0.55$ , $p < 0.05$ ), social anxiety ( $r = 0.64$ , $p < 0.05$ ), and the overall autism index ( $r = 0.56$ , $p < 0.05$ ).	10
Tudor et al (2012) <sup>68</sup> USA	Examined the relationships bet. sleep problems and ASD	ASD (n=109) M (83.49%), aged 3–18 M = 7.06 SD = 2.67	Independent pediatricians and psychologists	Cross-Sectional. Trained staff visited home for 1.5 hr. assisted mothers to complete questionnaires.	GARS, CSHQ.	Sleep onset delay was strongest predictor of communication deficit, stereotyped behavior, and autism severity. The parasomnias sub-scale significantly correlated with communication ( $r = 0.28$ , p < 0.055), social Interaction ( $r = 0.21$ , p < 0.05). Social interaction significantly correlated with Night waking ( $r = 0.26$ , p < 0.01) and sleep-disordered breathing ( $r = 0.19$ , p < 0.05).	12

Tyagi et al (2018) <sup>16</sup> India	Explored sleep problems and their association with behaviors, and comorbidities.	ASD (n=74) M (82%), aged 3–12 M=4.8; SD=1.8	SCQ, M-CHAT-R/F	Cross-Sectional. Excluded if on medications affecting sleep or chronic illness.	DP-3, The Conners ADHD Rating Scale-3, VABS-II, SDSC.	Sleep problems significantly associated hyperactivity (OR = 10.2, 95% CI 3.34–31.2, $p < 0.001$ ) and internalizing (OR = 1.09 95% CI 1.02–1.156, $p = 0.009$ ) and externalizing (OR = 1.078 95% CI 1.02–1.13, $p = 0.004$ ) behavior.	
Veatch et al (2017) <sup>20</sup> USA	Examined relationship bet. shorter sleep duration and ASD symptomatology	ASD (n=2713) M (86%), aged 4–18 M=9.04 SD=3.58	DSM V, ADI- R, ADOS	Cross-Sectional.  Data analyzed from Simons Simplex Collection.	ADI-R, ADOS, CBCL.	Social/ communication impairment ADI-R negatively correlated with parent-reported sleep duration ( $p=-0.19$ , $p=1\times10^{-5}$ ). Shorter sleep duration moderately correlated with Increased social affect scores ( $p=-0.06$ , $p=1.8\times10^{-2}$ ). Short duration was associated with hyperactivity (OR = 2.90, 95% CI = 1.66, 5.06, $p<=1.0\times10^{-4}$ ).	12
Wang et al (2016) <sup>69</sup> China	Characterized sleep disturbances and examined associated behavioral factors.	ASD (n=60) M (83.3%), aged 6 - 17 M = 11.53 SD = 2.92	DSM IV	Cross-Sectional. Participants recruited from a school for children with special needs.	SDQ, CSHQ.	Total CSHQ correlated significantly with total SDQ (r = 0.51, p < 0.001), hyperactivity (r = 0.47, p < 0.001), emotional symptoms (r = 0.34, p = 0.008), conduct problems (r = 0.38, $p$ = 0.003).	9
Yang et al (2018) <sup>70</sup> China	Examined association. bet. gastrointestinal, sleep problems and behavior and risk factors.	ASD (n=169) M (85.8%), aged 3 -12 M = 8.6 SD = 4.2	DSM IV, ADI- R, ADOS	Cross-Sectional.  Research Center of Harbin Medical University.	ABC, VABS, PDDSQ, SRS, CSHQ, VABS, PPVT, CARS, CABS	Children with and without sleep disturbances significantly differed in socialization item scores of VABS (p = 0.004), ABC scores PDDSQ scores, SRS total scores, social cognition and social Communication item scores of SRS respectively.	10
Yavuz-Kodat et al (2020) <sup>71</sup> France	Determined the relative contribution of sleep and circadian rest–activity rhythm to problem behaviors.	ASD (n=52) M (78.85%), aged 3–10 M = 5.39 SD = 1.5	ADOS, ADI- R	Cross-Sectional. Children included regardless of dev. delay or ID. Excluded if on unstable medication and if traveled over 2 time zones in preceding 3 months.	ABC-C, VABS, CSHQ, Actigraphy, NPCRA using rest-activity data.	The higher irritability and stereotypic behaviors group slept continuously (-60 min, $p = 0.04$ ) and (-75 min, $p = 0.006$ ) respectively than those with less irritability and stereotypic behaviors.	12

Abbreviations: ABC, Aberrant Behavior Checklist; ABC-C, Aberrant Behavior Checklist; ABC-C, Aberrant Behavior Checklist; ADI-R, Autism Diagnostic Interview, Revised; ADOS, Autism Diagnostic Observation Schedule; ASSQ, Autism Spectrum Screening Questionnaire; ATA, Assessment of Autistic Behavior; AuBc, Autism Behavior Checklist; BRQ, Bedtime Routines Questionnaire; BISQ, the Brief Infant Sleep Questionnaire; ICSD BEDS, a sleep problem questionnaire constructed from items from the International Classification of Sleep-disorders; CABS, Clancy Autism Behavior Scale; CARS, Childrond Autism Rating Scale; CBCL, Child Behavior Checklist Score; CSHQ, Childronds Sleep Habits Questionnaire; CSHS, Childronds Sleep Hygiene Scale; CSWS, Childronds Sleep-Wake Scale; DISCD, Diagnostic Interview for Social & Communication Disorders; DP3, Development Profile 3; DSM IV, Fourth Edition criteria; ES, effect size; HRQoL, Health-Related Quality of Life; IBQ-R, the Infant Behavior Questionnaire—Revised; M-CHAT-R/F, Modified Checklist for Autism in Toddlers, Revised; NPCRA, Non-Parametric Circadian Rhythm Analysis; NRI-RQV, Network of Relationships Inventory-Relationship Qualities Version; PCQ, Parental Concerns Questionnaire; PDDSQ, Pervasive Developmental Disorders Screening Questionnaire; PPVT-C, Peabody Picture Vocabulary Test-chinese edition; PSI, Parenting stress index-short form; PSQI, Pictsburgh Sleep Quality Index; PSQ, Paediatric sleep questionnaire; PedSQL 4.0, Pediatric Quality of Life Inventory 4.0; PSES, Percent Superiority Effect Size; RBQ-2, Repetitive Behavior Questionnaire; PGSQL, Social Communication Questionnaire; PGSQL, Social Communication Questionnaire; PGSQL, Social Polity Behavior Scale-Second Edition; ABA, Applied Behavioral Analysis; ASD, Autism Syndrome Disorder; Bet., between; Cog., cognitive; Dev., developmental; EEG, electroencephalogram; ICD-10, International Classification of Diseases; ID, intellectual disability; PSG, polysomnography; NT, neurotypical; TST, total sleep time; WASO, wake after sleep onse

Whelan et al **Dove**press

# Relationships Between Sleep Quality, Social Functioning and Behavior Problems **Externalizing Behavior Problems**

There is substantial evidence that externalizing behavior problems can be associated with sleep problems<sup>67</sup> and multiple hierarchical regressions have been used to show that they can be statistically predicted by sleep quality.<sup>59</sup> Fadini et al,<sup>39</sup> using parental reports, found sleep disorders, particularly arousal disorder and excessive daytime sleepiness, correlated with behavioral problems. This finding concurs with Aathira et al<sup>33</sup> which measured sleep prospectively with a small sample, using one night of PSG. This study found that poor sleepers had a mean Child Behavior Checklist Score of 63.8 (60.8 to 66.8) whereas good sleepers scored a mean of 57.8 (53.0 to 62.5)(P 1/4 0.004). Malhi et al<sup>52</sup> also found a significant relationship between sleep dysfunction and daytime behavior difficulties (r = 0.53, P = 0.01), and autistic children with sleep problems (t = 2.42, P = 0.018) had significantly more behavioral difficulties (M = 15.67, SD = 6.10) than those without sleep problems (M = 9.50, SD = 3.01). Several studies have found that sleep problems are significantly associated with irritability, hyperactivity, inattentiveness, 20,41,69,71 and aggression, which parents attributed to tiredness. 48 Sleep disturbance has also been identified as a predictor of attention deficit, hyperactivity, and oppositional defiant behavior.<sup>47</sup> Allik et al<sup>27</sup> found that 31.2% of their sample of autistic children, without ID, had insomnia. These children had more teacher-reported hyperactivity symptoms than those without insomnia. Goldman et al<sup>42</sup> also found that the poor sleeper group within their sample showed more inattention and hyperactivity, and they had higher scores on compulsive and ritualistic scales in comparison to the good sleeper group. Goldman et al<sup>43</sup> also found that 50% of the poor sleeper group had problems with attention, social interaction, language, hyperactivity, sensory issues, eating behaviors, and self-stimulatory behavior, in comparison 50% of the good sleeper group who only reported problems in language, attention, and social interaction. The characteristics of good sleepers versus poor sleepers have also been examined in an RCT. 28 In this RCT, the poor sleeper group had significantly higher daytime behavioral problems including, hyperactivity, stereotypical behaviors, and more irritability. However, Lambert et al<sup>4</sup> found that SWS positively correlated with the score of the ADI-R subscale for repetitive behavior (r = 0.714, p = 0.02), a positive association was difficult to explain.

### Internalizing Behavior Problems

Sleep disturbances have been associated with anxiety<sup>43</sup> and emotional problems.<sup>27,46,47,69</sup> Lambert et al<sup>50</sup> used data collected through parent-reported questionnaires, daily sleep diaries, and laboratory PSG (2 nights). Their results showed that the amount of slow-wave sleep (SWS) correlated negatively with the amount of Internalizing behaviors (r = -0.41, p = 0.046), and the reciprocal social interaction score ADOS (r = -0.807, p = 0.003). Sleep latency, which was defined as the first occurrence of 10 consecutive minutes of stage 1 sleep or the first epoch of any other sleep stage, and SWS were associated with low levels of internalizing behavior and the social functioning of children as determined through ADOS measurements. Poor sleep quality is also associated with lower mood.<sup>27</sup> Kirkpatrick et al<sup>48</sup> examined the impact of insomnia, using focus groups (n = 2) that involved family members (n = 15). Their findings revealed that 26% percent of parents said that tiredness negatively impacted the mood of their children. Some evidence suggests that lower sleep quality may lead to poor prosocial behavior and social withdrawal. <sup>28,69,71</sup> One study examined the association between sleep problems and the quality of peer relationships. 62 This involved a small sample of verbally fluent autistic adolescents (n = 19) that measured seven nights' sleep using actigraphy. The questionnaires, but not the actigraph data indicated significant associations between sleep problems and discordant peer relationships (0.59 p < 0.01). Participants with more sleep problems reported discord with peers but the closeness of peer relationships was not significantly associated with sleep quality. They concluded that autistic adolescents can be vulnerable to negativity in peer relationships when they experience sleep problems and that they may have difficulty regulating social interaction.

In one study, social withdrawal, and stereotypic behaviors accounted for 17% and 36% of the variance, respectively, in sleep disturbances.<sup>71</sup> It has also been suggested that the need for social interaction can decrease with increased sleep problems.67

#### Aspects of Sleep Quality

Particular aspects of sleep quality including sleep onset delay/sleep latency, short sleep duration, and shorter continuous sleep have been associated with social functioning. For example, Tudor et al found a positive correlation between stereotyped behavior with sleep onset delay (0.31 p < 0.01), and social interaction (0.40 p < 0.01). Sleep duration also correlated with stereotyped behavior (0.32 p < 0.01) and social Interaction (0.33 p < 0.01). Also, when controlling for the variance contributed by other sleep problems, sleep onset delay (pr = 0.27, p < 0.01) and night wakings (pr = 0.22, p < 0.05) were significantly strong predictors of stereotypical behaviors.  $^{68}$ 

There is some evidence that shorter sleep latency relates to poorer social functioning. Naito et al $^{58}$  examined the relationship between body movements at night and social ability, in a small sample of relatively young autistic children (n = 17) arguing that movement would correspond to periods of light rather than deep sleep. Dissimilar to previous studies that used PSG, they measured the body movement of participants sleeping in their own homes for 3 nights using an accelerometer attached to the waist. They found that a higher rate of body movement at 0.5 to 1 hour after onset body stillness was associated with lower social ability. They suggested that higher social symptoms severity was associated with shorter latency in the first nocturnal peak of movement and in the latter half of the night. This study did not evaluate motor function or measure the amount of daytime activity that may impact movements at night and they did not measure body movements for 5 nights, which is optimal to estimate body movement patterns.<sup>72</sup>

Other evidence indicates that the duration of uninterrupted continuous sleep is important. Delahaye (2014) found the largest correlations were between sleep duration and quality of life total (-0.36, p < 0.001), and psychosocial summary (-0.36, p < 0.001) scores, when they investigated the relationship between sleep problems and quality of life, child age, gender, autism diagnosis, internalizing and externalizing behavior, and autism severity. Furthermore, extremely short sleep duration increased core autism symptom severity<sup>22</sup> and a short duration of uninterrupted sleep may be more impactful than sleep latency. 65,71 These results concur with those of Yavuz-Kodat et al. 71 who examined the relationship between sleep, circadian rest-activity rhythm to problem behaviors. This study found children with shorter continuous sleep had high irritability in comparison with those with lower irritability (-60 min, p = 0.04), and high stereotypic behaviors compared to children with fewer stereotypies (-75 min, p = 0.006). Shorter continuous sleep was 60 minutes shorter in the higher irritability group and 75 minutes shorter in the higher stereotypy group. Yavuz-Kodat et al determined that children with shorter continuous sleep completed approximately one less complete sleep cycle of REM and non-REM sleep. Further evidence of the importance of uninterrupted sleep can be found in a study that evaluated the effects of slow-release melatonin treatment on sleep and behavior.<sup>65</sup> Schroder et al<sup>65</sup> found a modest correlation between the number of difficulties experienced by participants and increase in total sleep time from baseline (Spearman's rank correlation R - 0.229 p = 0.024). Schroder et al<sup>65</sup> attributed this to the increased duration of uninterrupted sleep (Spearman's rank correlation R - 0.21 p = 0.047) but not with the shortening of sleep latency (Spearman's rank correlation R 0.09 p = 0.379). Taking the slow-release melatonin drug resulted in the Ped- PRM-treated participants sleeping for an average of 57.5 minutes longer. The placebo group had an increase of 9.14 minutes longer. The sleep latency of the Ped- PRM-treated participants decreased by 39.6 minutes on average compared to 12.5 minutes with placebo.

#### **Evidence Suggesting Causal Relationships**

Studies that have examined the relationship between sleep quality and social functioning longitudinally or with social functioning and behavior problems as outcomes during the evaluation of sleep interventions provide evidence that suggests improving sleep quality may improve these outcomes. For example, Malow et al conducted an RCT that used parental reporting and no placebo to assess the response, tolerability, safety of a slow-release Melatonin. After 14 weeks, sleep latency significantly decreased and there were significant improvements in sleep duration, social withdrawal, affective, and ADHD subscales. Another double-blind RCT evaluated the efficacy of prolonged-release melatonin mini-tablets (PedPRM; 2–5 mg) in improving sleep onset, duration, and child behavior. Participants had either autism or Smith-Magenis syndrome (n = 125). After 13-weeks, melatonin effectively improved sleep quality and behavior in comparison to a placebo. There were statistically significant improvements in externalizing behavior, which decreased on average – 0.70 units in the Ped- PRM-treated (N = 54), compared to worsening with an increase

of 0.13 units in the placebo-treated group (N = 49). In addition, 53.7% of patients in the PedPRM group improved by 1 or more units on externalizing behavior score compared to 27.7% in the placebo group (Odds ratio 3.0; p = 0.008). However, there was no significant difference in the mean scores of attributes that related to Internalizing behaviors (peer relationships and emotional behavior). Overall, 47% of subjects had a reduction of 1 or more in total strengths and difficulties score in the PedPRM group, compared to 27.1% in the placebo group (p = 0.035). This suggests that hyperactivity/inattention may be relatively more responsive to the sleep improvements that result from treatment with melatonin than Internalizing behaviors. However, it is noteworthy that this study was powered to detect differences in effects on sleep but not behavior. Papadopoulos et al<sup>60</sup> conducted an RCT pilot study that evaluated the efficacy of a brief behavioral sleep intervention on sleep and child functioning. Participants who were children with an ADHD-Autism codiagnosis were randomized to the sleep intervention group (n = 28), or the group receiving usual care (n = 33). This study assessed the mean difference in change scores between baseline to 3 and 6 months between the groups. They found that the intervention group improved emotional functioning to a greater degree according to parent reports at 3 months: (mean difference = -1.0, 95% CI = [-2.0, -0.1]; ES = -0.6, p = 0.04), and teacher reports at 3 months (mean difference = -1.2, 95% CI = [-2.3, -0.1]; ES = -0.5, p = 0.04). However, by 6 months, there was no significant difference in emotional functioning, but teacher reports suggested there was an improvement in conduct problems. Small improvements in peer relationships at 6 months were detected by both parents and teachers (mean difference = -0.6, 95% CI = [-1.6, 0.4]; ES = -0.3, p = 0.22) and teacher report (mean difference = -1.0, 95% CI = [-2.3, 0.3]; ES = -0.4, p = 0.11). Papadopoulos et al<sup>60</sup> concluded that the intervention resulted in large improvements in sleep and moderate improvements in behavioral function at 3 and 6 months.

## Related Factors

## The Severity of Autism Core Symptoms

Some studies have examined social functioning focusing on the severity of autism symptoms. These studies provide mixed evidence concerning the relationship between sleep quality and the autism severity. The majority of studies have found that a positive association exists between them. <sup>20,22,37,64,67,68</sup> For example, Sare & Smith (2020), <sup>63</sup> in a study involving secondary data analysis of a large parent-reported data set (n = 19,104), found a strong association between sleep problems and the severity of autism-related behaviors. However, in two studies, sleep disturbance scores did not correlate with autism severity<sup>33,54</sup> and in another, a small amount of variance in daytime symptoms was explained by sleep problems.64

The evidence suggests that hyper-arousal and increased sensitivity to stimuli in the sleep environment are associated with sleep disturbance.<sup>54</sup> Manelis-Baram et al<sup>54</sup> found that the sensory sensitivity score exhibited the strongest correlation with sleep duration, and sleep disturbances correlated with sensory problems correlated (r = 0.57, p < 0.001), sensation avoiding (r = 0.49, p < 0.001), low registration (r = 0.37, p < 0.001), and sensation seeking (r = 0.24, p = 0.014) scores. Further evidence of the relationship between sensitivity and sleep problems was obtained through a study that found significant associations between sleep problems and increased seeking, touch, movement processing patterns, conduct, and attentional responses to sensory processing.<sup>20</sup> Mazurek et al<sup>55</sup> also found sleep problems correlated with sensory over-responsivity, and the study results suggested that sensory over-responsivity was a longitudinal predictor of sleep problems and inattention/hyperactivity in very young, but not older children.

#### Cognitive Ability

Several studies found that the relationship between sleep quality and social functioning exists regardless of cognitive ability and the presence/absence of ID. 28,41,47,52,54,63 In contrast, a small number of studies found that the quantity and quality of sleep are associated with cognitive performance<sup>50</sup> and that the presence of ID was an important predictor in the relationship between sleep quality and emotional and behavioral disturbance. 47 Krakowiak et al 49 also found, in a large population-based study, that lower cognitive and adaptive function is associated with increased delayed sleep onset, night awakening, and shorter sleep duration. In addition, two other studies found that individuals with an unstable sleep phenotype had a relatively lower IQ and adaptive functioning level than those with a stable phenotype. <sup>37,38</sup>

#### **Anxiety**

There is some evidence that anxiety contributes to the association between sleep and social functioning. 21,46,47,57,68 However, Mazurek et al's<sup>55</sup> results did not indicate a significant predictive relationship between anxiety and sleep problems after accounting for covariates, even though anxiety correlated with sleep in both older and younger children. However, a causal relationship between anxiety and sleep quality was described by eighty-six percent of parents (n = 13) in one study.<sup>48</sup> Parents said that their children experienced sleep-related anxiety that resulted in delayed bedtimes, unwanted co-sleeping, night awakening disruptive night-time behaviors. 48 They reported that children were afraid to sleep alone, anxious about darkness, worried about forthcoming events or school and that this anxiety contributed to reduced and poor quality sleep. Therefore, anxiety may impact sleep quality by delaying sleep onset and pre-bedtimes routines.<sup>44</sup> Indeed, the consistency of pre-bedtime routines can be negatively associated with externalizing behaviors (b = 0.55, t(110) = 6.76, p < 0.001) and sleep quality. The consistency of routines is related to problematic pre-bedtime behavior and bedtime resistance<sup>40,48</sup> and children with higher irritability have more difficulties regarding bedtime routine. 41,44,71 Goldman et al 42 also found that this was a major concern by parents of children who were categorized as poor sleepers. Bedtime resistance was also identified as a risk factor for repetitive behavior in preschool autistic children and it explained a large proportion of variance in repetitive behavior scores (adjusted R2 = 0.206). 46 In addition, linear regression showed that bedtime resistance and sleep onset delay explained a large and considerable proportion of the variance in the total repetitive behavior score (20.6% and 17.3%), respectively.<sup>46</sup>

#### Age

As suggested above, the relationship between sleep quality and social functioning may differ according to age and behavior problems and a later sleep midpoint of older children was associated with challenging behavior.<sup>34</sup> Veatch et al<sup>20</sup> evaluated the effects of age on associations of sleep duration and autism traits in children who were 4–5 years old (17.7%); primary school age (63.6%), and secondary school age (18.5%). This study found that age is significantly related to short sleep duration, anxiety, depression, Internalizing problems, and the severity scores for affective problems. Goldman et al<sup>43</sup> also found an association between parasomnias and behavior across the age span with an inverse relationship between age and behavior problems. However, in contrast, Johnson et al<sup>28</sup> found no age difference between good sleepers and poor sleepers in terms of disruptive behaviors, and parasomnias have been found to persist across an age range of 3–18 years.<sup>43</sup> Furthermore, even adjusted for age, poor sleepers were found to be at the highest risk of self-injurious behaviors.<sup>34</sup>

#### Temporal Aspects

A few studies have examined the relationship between sleep and functioning over time. May et al<sup>21</sup> investigated the bidirectional relationship between sleep disturbance and behavioral, and emotional problems, longitudinally, at baseline (T1) and 1 year later (T2). At both time points, sleep problems correlated with social difficulties, aggression, and hyperactivity. These results concurred with Mazurek et al,<sup>55</sup> who examined a large well-characterized sample of autistic children at baseline (T1) and M = 3.8 years later (T2). Mazurek et al found that sleep problems were significantly associated with most co-occurring symptoms at T1 and T2.

For individuals over time, the evidence varies as to whether the relationship between the trajectories of sleep problems and social functioning can persist over time. They can be persistent, but children with greater sleep problems at the second time point (aged 4) had higher trajectories for repetitive behaviors. <sup>47</sup> In addition, several studies identified a high degree of individual variability. <sup>34,37,40</sup>

It is likely that the chronicity of disturbed sleep may have an accumulative effect on social functioning. Abel et al<sup>34</sup> used objective measures of sleep and behavior to explore night-to-night fluctuations in sleep patterns and challenging behavior, ie, whether challenging behavior would follow the day after a poor quality night's sleep. This study found, using multilevel models for predictive analysis, that across eight nights, rather than a single night, sleep quality predicted the subsequent challenging behavior. This finding concurred with Cohen et al,<sup>38</sup> who examined data from five days/nights measurement of sleep and challenging behaviors. They collected sleep data using structured observations and clinicians worked one on one with participants to observe and record behavior hourly each day for up to 21 hours. They found that

Whelan et al Dovepress

the previous night's sleep did not strongly predict next-day problem behaviors. Instead, behavior was affected after several nights of poor sleep, and the accuracy of predicting behavior increased with the number of nights measured. This finding concurs with qualitative research in which a participant parent said planned family trips to the cinema were canceled after: "a few late nights and early mornings. Well! He [the autistic child] can be awkward and it's just not worth it" (Kirkpatrick et al, 2019, Parent 13 p. 64).<sup>48</sup>

## **Discussion**

# Summary of Key Findings

The evidence presented in this review reveals that the social functioning and problem behaviors of autistic children, with or without ID, are associated with their sleep quality. A relatively small number of studies have evaluated social function and behavior problems as outcomes of sleep interventions, but these reveal evidence that suggests causal relationships and that social functioning may improve as sleep improves but that the improvements may be temporary. 53,60,65

Social functioning is associated with the tendency for autistic children to have shorter latency in the first nocturnal peak, less deep, and more fragmented sleep than those with NT.<sup>58</sup> Therefore, the relationship between sleep quality and social functioning is probably unique to autistic individuals.<sup>68</sup>

More specifically, chronic and accumulative poor sleep quality can increase externalizing behavior problems including hyperactivity and inattentiveness. However, a few of the study samples examined contained autistic participants who were also diagnosed with ADHD, and the majority of studies did not out rule the presence of ADHD. Therefore, definitive conclusions cannot be reliably drawn due to the high comorbidity rate between autism and ADHD.<sup>73</sup> The evidence also reveals that poor sleep is negatively associated with problematic Internalizing behavior, increased negative mood, and social withdrawal. However, further research needs to investigate the impact of sleep on Internalizing behavior as discussed below.

Overall, the evidence suggests that anxiety negatively impacts sleep onset, duration, and it increases sleep disturbance. Therefore, the relationship between sleep quality and social functioning is bi-directional. This is an important finding as anxiety disorders are common in autistic individuals with prevalence rates from 42% to 79%. <sup>74</sup> Related to anxiety, hyper-arousal and sensitivity are associated with sleep disturbance. <sup>20,54,55</sup> Hyper-arousal has also been associated with insomnia in autistic adults. <sup>75</sup> This finding also supports the presence of a bi-directional relationship between sleep quality and social functioning in autistic children and it supports the hypothesis that particularly in young children, sleep disturbance and sensory sensitivities are likely to be co-dependent, and generated by interacting physiological mechanisms. <sup>54</sup>

There is mixed evidence regarding the relationship between sleep quality and social functioning and its association with autism severity and age. On balance, the evidence reveals an association with the severity of autism symptoms, and that the relationship changes according to age. Previous related research has identified that autistic individuals have age-related variations in core symptom severity<sup>76</sup> and sleep behavior.<sup>73</sup>

The majority of studies reviewed used a cross-sectional research design and a small proportion were longitudinal. <sup>21,34,40,54–56,60</sup> Therefore, there is little evidence with which to determine causality and its direction with any degree of certainty. It is also notable that only one study employed qualitative research methodology. <sup>48</sup> Most studies had relatively small sample sizes. However, there were a few large population-based studies. <sup>22,43,49</sup>

Examining the relationship between sleep quality and social functioning is complex as it is impacted by comorbidities that are common and experienced by up to 46% of autistic individuals. 38,77 The studies reviewed varied hugely regarding which confounding factors researchers controlled for or excluded through participant selection. As discussed above, studies did not all exclude children with co-diagnosis of ADHD, and only some controlled for medical comorbidities, 46,53 and pharmacological agents. There was a similar variety regarding other confounding factors. For example, few studies assessed the impact of daytime sleep or daytime activities and none recorded whether participants slept in the same room as other children and if the latter had sleep problems. Kelmanson et al 47 were an exception in recruiting only singletons without severe comorbidities.

Most of the studies relied on parental reporting for measuring sleep and/or behavior although three studies assessed daytime social functioning with teacher/caregivers reports that were blinded to the sleep quality. <sup>37,38,60</sup> The use of parental reporting means most assessments were unblinded and were therefore, subject to reporter bias. Findings using parent-reported sleep measures can concur with those from objective measures. <sup>20,23,27,42</sup> But, they do not always do so, <sup>35</sup> and sleep duration is the most reliable sleep trait reported by parents. <sup>22</sup> Furthermore, the use of parental measures for behavior, and not including reports from child/adolescent participants, is likely to underestimate Internalizing behavior, emotion, anxiety, and feelings of sleepiness. <sup>65,67</sup> Notably, only two studies in this review collected data directly from autistic adolescents. <sup>62,67</sup> Underestimation of Internalizing behavior is concerning particularly as self-injurious behavior is a high risk for poor sleepers of all ages <sup>43</sup> and worry/rumination can predict the later development of Internalizing and externalizing problems in autistic children. <sup>78</sup> But, it may be more problematic for adolescents because the reported somatic complaints of individuals increase over time with age. <sup>78</sup> There are methodological challenges in involving people with autism who have an ID but these can be minimized to facilitate their participation in future research. <sup>79</sup>

Only one study measured both sleep and social functioning objectively,<sup>34</sup> and two related studies used structured sleep and behavior observations.<sup>37,38</sup> The use of objective methods potentially reduces bias and several studies measured sleep objectively using actigraphy, 20,36-38,40,53,58,62,80 and PSG. 33,42,50 Moore et al 81 reviewed the literature and reported that actigraphy can underestimate SOL in autistic children, but that there is good compliance with this method and minimal data loss. The validity of actigraphy against PSG has been assessed in autistic children (n = 26) during one night in a hospital setting.<sup>82</sup> This study found good agreement between measures for SOL, TST, and moderate agreement regarding WASO and SE. However, in this review, there was a large variation regarding whether sleep diaries were used to provide contextual data, how children and parents were prepared for PSG and actigraphy, and regarding the duration of the measurement. These are important considerations for the validity of measurements because usual sleep behavior may be distorted if it is measured in ways that are challenging for autistic children who have sensory issues and a preference for sameness.<sup>3</sup> PSG may be challenging because it involves sensors being attached to the face and scalp, and it requires children to sleep in an unfamiliar environment laboratory environment. Malow et al<sup>83</sup> found that autistic children who were poor sleepers had a significantly poorer SOL and SE in comparison to autistic good sleepers and NT control groups measured on one night by PSG. However, after subsequent nights of PSG measurement, there was less difference between the groups. Malow et al<sup>83</sup> recommended that the validity of PSG can increase if autistic children are acclimatized to PSG for two nights and then have their sleep measured during a third night in the laboratory.

## Future Research

Future research needs to explore further the impact of confounding variables on the relationship between sleep quality and social functioning. Doing so could be helpful through explaining the variability between individual children. <sup>34,37,40</sup> For example, the onset of puberty may be a potential confounder <sup>22</sup> and this has not been investigated to date. Other factors, including gastrointestinal symptoms, <sup>70</sup> dietary and eating habits, <sup>48</sup> and breathing problems, <sup>59</sup> have been afforded minimal attention. Future work should also clarify how gender impacts sleep quality and social functioning. Except for May et al, <sup>21</sup> whose sample had a more equal gender ratio, the reviewed studies contained samples of predominantly males, as is typical within the autism population. <sup>2</sup>

Sleep quality and social functioning are both impacted by a mixture of internal and external factors, including interpersonal interactions.<sup>38</sup> They are also associated with wider societal policies and cultural practices.<sup>47</sup> To date, research has focused on issues that are personal to and within individual autistic children. Future research should explore the impact of wider socio-cultural factors in general, and specifically parental stress, the parent–child relationship, and co-sleeping practices. It can be comparatively more stressful to parent an autistic child than an NT child<sup>84</sup> and this may be aggravated through a child's poor sleep quality.<sup>48</sup> The use of mixed methods/qualitative research methodology may facilitate in-depth exploration of potential environmental and societal issues.

Sleep quality and social functioning have predominantly been studied using cross-sectional cohorts with data collected at one point in time. More longitudinal and prospective research are required to confirm causal relationships. These designs are also necessary to increase understanding of the trajectory over time and age. Knowledge from this research will facilitate clinicians to offer individualized interventions.

Whelan et al Dovepress

Future research using Delphi survey techniques<sup>85</sup> would also be helpful to increase consensus regarding the management of confounding factors in future research and to facilitate the development of best practice guidelines regarding the use of actigraphy and PSG with autistic individuals.

# Implications of Findings

Because autistic children may have poor quality sleep and not complain about this,<sup>50</sup> all parents of autistic children may benefit from receiving information that aims to increase sleep quality, because this is likely to maximize their child's social functioning potential. Also, sleep quality should be assessed for autistic children who present to services with behavioral problems and sleep interventions should be integrated into treatment plans.<sup>27,52,57,59</sup>

# Limitations and Strengths

This review used systematic and robust methods and the methodological strengths and weaknesses of studies were critically appraised using a recognized tool. However, the review only included studies published in English that focused on sleep quality, social functioning, and behavior problems. Therefore, some studies could have been excluded that might have been informative, particularly those that focused on related concepts including communication, and quality of life.

## **Conclusions**

This review examined 46 studies to elucidate understanding of the relationship between sleep quality, social functioning, and behavior problems. Evidence revealed that poor sleep quality is associated with social functioning and behavior problems. A small amount of evidence suggests the existence of a bi-directional causal relationship between sleep quality and social functioning. Delayed sleep onset, reduced sleep duration, and fragmented sleep without continuous periods of uninterrupted sleep, over several days, appear to increase problematic externalizing and Internalizing behavior. However, anxiety, hyper-arousal, and sensitivity may negatively impact sleep quality. RCTs and prospective studies are required to establish causality. Future research also needs to further examine confounding factors, increase consensus for the objective measurement of sleep with autistic individuals, explore the impact of socio-cultural practices, and increase understanding about the Internalizing behavioral consequences of poor sleep quality.

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Whelan et al Dovepress

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