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## **Sleep and cognition in people with Autism Spectrum Condition: A systematic literature review**

### **Running header: Sleep and cognition in people with ASC**

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### **Conflict of Interest**

The authors have no conflicts of interest of declare.

**This work** has not been published before and it is not under consideration for publication elsewhere.

## Abstract

Sleep disturbances have been found to increase severity of core Autism Spectrum Disorders (ASD) symptoms such as social and communication difficulties and repetitive behaviours. However, very little is known about the impact of sleep on cognitive functioning in ASD. A systematic literature search identified fifteen original studies meeting inclusion and quality assessment criteria. Inter-rater reliability was strong ( $\text{Kappa}=.73$ ). Multiple cognitive functions were negatively affected by sleep disturbances in ASD. This had a significant impact on daily functioning, as well as impacting families/carers. Possible sleep interventions include behavioural/cognitive training, pharmacological treatment, and sensory interventions. Combining objective and subjective sleep measures would help in early diagnosis, which may contribute towards developing interventions to help those with ASD as well as carers and families.

Keywords: Autism Spectrum Disorders, sleep, cognitive functioning, systematic literature review

## **Sleep and Cognition in People with Autism Spectrum Disorders: A Systematic Literature Review**

The DSM 5 (APA, 2013, P. 299F84) defines Autism Spectrum Disorder (ASD) as “characterised by persistent deficits in social communication and interaction, as well as restricted, repetitive patterns of behaviour, activities or interests”. Sleep disturbances are common amongst people with ASD, affecting around 40-80% of children (Cohen et al., 2014) compared with 9% to 50% among typically developing (TD) children (Park et al., 2012), and also affecting 80% of adolescents and young adults (Oyane & Bjorvatn, 2005). Sleep disturbances encompass a range of problems, including difficulty initiating and/or maintaining sleep, difficulty achieving restorative or satisfactory sleep, with possible associated daytime functioning impairment, as well as clinically diagnosed sleep disorders (Edinger et al., 2004; Ohayon, 2002). The following review will use the term ‘sleep disturbances’ as an umbrella term for all known sleep problems found in people with ASD.

Functional magnetic resonance imaging (fMRI) studies comparing sleep-deprived and well-rested brains provide significant evidence that sleep is essential for optimal cognitive function and learning (Chee & Chuah, 2008). Cognitive functioning is an intellectual process which involves being aware, perceiving and comprehending ideas. It encompasses all facets of thinking, reasoning, perception and memory (Gazzaniga, Ivry, & Mangun, 2009). For example, in a randomised study examining the effects of sleep deprivation in medical students, those who worked a traditional schedule were found to make 36% more medical errors than the medical students who worked under an intervention schedule which included more sleep (Landrigan et al., 2004). Sleep disturbances can also have a severe impact on an individuals’ ability to learn new skills (Chee & Chuah, 2008). The impairments that result from sleep disturbances comprise diminished cognitive and behavioural functioning, partly

from reduced attention and arousal. In addition, cognitive processes such as memory consolidation and insight formation occur during sleep and are essential elements of knowledge acquisition (Ellenbogen, 2005).

In individuals with ASD, studies have shown that sleep disturbances can lead to an increase in the severity of core ASD symptoms such as social and communication difficulties and repetitive behaviours (Park et al., 2012; Tudor, Hoffman, & Sweeney, 2012). Sleep onset and maintenance problems and short sleep duration are consistently those of highest concern to parents of children with ASD (Richdale & Schreck, 2009). Park and colleagues (2012) found that children with ASD were more likely to experience bedtime resistance, insomnia, and daytime sleepiness compared to their unaffected siblings, and that sleep problems were significantly associated with co-occurring psychopathology such as withdrawal problems, aggressive behaviours, internalising problems, and total behavioural problems. The severity of sleep problems in children with ASD is a matter of concern due to increased stress experienced in parenting a child with ASD and the adverse effects of sleep disturbances on daytime behaviour and general functioning of the child (Hill et al., 2014; Limoges et al., 2013; Park et al., 2012).

### **Rationale for review**

A recent review of the literature examined the association between sleep disturbances and its impact on the problematic behaviour of people with ASD (Cohen et al., 2014). Cohen and colleagues concluded that although sleep interventions might help with problematic behaviours, there is a gap in the literature for understanding the broader impact of sleep disturbances. In typical developing children, sleep disturbances have been shown to have a negative impact on cognitive functioning (Chee & Chuah, 2008) however little is known

about the impact of sleep disturbances on the cognitive functioning in people with ASD. For some individuals, this could be due to communication difficulties being a core symptom of ASD, and as such, they struggle to report their sleep disturbances (Cohen et al., 2014; Limoges et al., 2013). Hollway et al. (2013) found a bidirectional association between sleep disturbances and severity of ASD symptoms (see also Park et al., 2012; Tudor et al., 2012). Unlike in typically developing children, sleep difficulties in children with ASD appear to be chronic and continue throughout their lifespan (Cohen et al., 2014). Therefore, there is a need to review how sleep affects cognitive functioning in people of all ages with an ASD.

### **Aims and objectives**

This systematic review aims to identify and critically evaluate available research that looks at how sleep affects cognitive functioning in people with ASD. This understanding may lead to developing clinical interventions for managing sleep disturbances in a person with ASD and/or their parents. This review aims to address the following questions:

- 1) How do sleep disturbances impact on the cognitive functioning of people with ASD?
- 2) What are the principal challenges associated with studying the impact of sleep disturbances in people with ASD?

## **Methods**

### **Literature Search**

A systematic search of the literature was conducted for qualitative and quantitative studies that have investigated sleep problems and their effect on cognitive functioning in people with

ASD. The studies included in this search were from 1994 onwards to include participants that had been diagnosed with ASD from the DSM-IV & DSM-5 criteria. Due to the difference in diagnostic criteria of ASD across the world only countries that use the DSM were included (Elsabbagh et al., 2012). Asperger's syndrome (AS) was a separate diagnosis to ASD prior to DSM-5; therefore earlier papers may refer to AS whilst later papers do not.

This systematic review followed the reporting guidelines and criteria of Preferred Reporting Items for Systematic Reviews (Moher et al., 2009). Articles were searched using seven electronic databases: PsychINFO, Medline, Scopus, ScienceDirect, SAGE, Cochrane Library, Web of Science and Google Scholar. The reference lists of extracted articles were examined for additional relevant articles. Searches were also carried out using non-electronic sources such as the library book catalogues. Finally, unpublished works were searched via Google search, Coventry University Locate, and The University of Warwick Online Library search. Ethical approval was obtained from Coventry University Research Ethics Committee. The study also adhered to the BPS Code of Ethics and Conduct (British Psychological Society, 2009) and the Code of Human Research Ethics (British Psychological Society, 2010).

### **Search terms and strategy**

Key search terms include the main concepts and synonyms, located within the title, abstract or keywords. For instance, when searching for the main concept *cognitive functioning* synonyms such as cognition were also accepted. The search strategy involved: Sleep AND Autis\* OR ASD OR Autism Spectrum Disorder AND Cognitive functioning OR Cognition.

### **Selection criteria**

Article titles and abstracts were screened and retained if they were written in English, were peer reviewed qualitative or quantitative empirical studies and were fully accessible. The study selection process is illustrated in Figure 1. In total 338 articles were initially identified. Of these, a total of 15 studies met the criteria for inclusion in this systematic review. Further information on each of these studies can be found in the supplementary material.

### **Inclusion Criteria**

All the articles were from 1994 onwards. Only those studies that originated from developed countries of UK, USA, Canada, Europe, Australia and New Zealand were included. In terms of the participants, the inclusion criteria included both male and females with ASD of all ages, so included studies on infants, children, and adults with ASD.

### **Exclusion Criteria**

Research excluded from the review included studies that were not published in English and those prior to 1994. If the study provided a Title or an Abstract only these were also excluded due to lack of information. Ten studies that included no measures of cognitive functioning, and 195 studies that did not relate to sleep research, were excluded. Seven studies (e.g. reviews or commentaries) were also excluded as they were not considered full empirical research articles. A further five articles were removed due to their geographical location, i.e., these were studies conducted outside those countries stated in the Inclusion Criteria. Another nine full-text articles were excluded as their topics were not relevant to the subject area of the review (i.e., sleep and cognition in ASD).

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Figure 1 here

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### **Standard Quality Assessment Criteria**

Standard Quality Assessment Criteria (SQAC) was used to evaluate the quality of the 15 studies from the study selection process (Kmet, Lee, & Cook, 2011). The SQAC was used as it provides a quantitative, reproducible and systematic means of assessing the quality of quantitative and qualitative study designs, and it has been shown to be effective in assessing the quality of research in other systematic literature reviews of sleep (Henry et al., 2016; Koffel et al., 2015; Miller et al., 2014). No qualitative studies were included; thus, only the quantitative checklist was utilised.

The quantitative checklist contains 14 questions, which are assessed as the item being completely met (Yes: 2 points), partially met (Partial: 1 point), not met or not applicable (No or n/a: 0 points) (Kmet et al., 2011). Each study under review was scored using the checklist, and a percentage score calculated. Kmet et al. (2011) suggest a cutoff score of >50% for inclusion. As all 15 studies scored above 50% using the SQAC, no study was removed from the review (see Table 1). To improve the reliability of the SQAC, 50% of the studies were assessed by another researcher and an inter-rater reliability analysis using the Kappa statistic was performed. The second rater was independent of this review. An overall Kappa score of .73 indicated strong inter-rater reliability, with no individual study falling below .51.

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Table 1 here  
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## **Results**

### **Characteristics of Studies**

A summary of the key characteristics of the 15 studies included in this review can be found in the supplementary material. Geographically, two studies were from the UK; seven from the USA, one from Canada, two from Australia and three from Italy. Five studies were randomised controlled trials, four studies were a cross-sectional design, three a correlational design, one an exploratory design, one an experimental design and one a case-control design. No qualitative studies were found. The age of participants varied among the studies; 11 studied participants <19 years; two studied participants >16 years; two were not specific about participant ages. The sample sizes ranged from 14 to 1859 participants.

The majority of the reviewed studies used only subjective measures of sleep such as sleep questionnaires and sleep diaries, as well as a battery of questionnaires to measure the outcomes of cognitive functioning in the participants, such as the Pediatric Quality of Life Inventory (PedsQL) or the Wechsler Adult Intelligence Scale (WAIS-III). Only five studies included objective measures of sleep such as actigraphy and polysomnography in addition to subjective measures. Further, two studies undertook patient histories. One of these two studies conducted additional clinical examinations and neuroimaging tests (e.g. CT, MRI scans) to collect neurophysiological data in the ASD participants.

### **How do sleep disturbances affect cognitive functions in people with ASD?**

Four of the 15 studies explicitly discussed cognitive functions affected by a disruption of sleep in individuals with ASD (Bruni et al., 2007; Limoges et al., 2013; Maski et al., 2015; Taylor et al., 2012). For instance, Bruni et al. (2007) offered a series of answers for why restorative sleep improves cognitive performance. Sleep questionnaires, neuropsychological tests, and polysomnography (PSG) recordings were employed to collect data for the study. The authors hypothesised that a high level of slow oscillatory activity during sleep is associated with improved performance in prefrontal cortical tasks. Oscillatory activity is the recurring and rhythmic electrical activity produced spontaneously as a reaction to stimuli supplied by neurons in the central nervous system (Başar, 2013). It aids the long-term consolidation of new memories during sleep; thus, an increase in slow oscillations enables more potentiation which then provides better cognitive performance. The results indicated that children with AS showed an increase in slow oscillatory activity during sleep, which could help to explain why some people with AS have superior memory abilities and are exceptionally talented or skilled in one or more specific areas. In addition, parents of children with AS reported more problems with daytime sleepiness and initiating sleep compared to those of TD children. We scored the Bruni et al.'s (2007) study high on the SQAC score (22/28=78.6) as this study was of good quality and provided detailed methodology. The study would have been scored higher if a larger sample size was included as the study only had eight children with AS and 10 with autism.

Similar to Bruni et al.'s (2007) findings, parents of children with ASD in Maski et al.'s (2015) study also reported more sleep problems than parents of TD children. Maski and colleagues studied 22 children with ASD and 20 control participants between the ages of nine and

sixteen years, and examined the role of sleep in declarative memory. Declarative memory is a type of memory dealing with everyday events and factual knowledge, and is dependent on the medial temporal cortex (Squire 1992, 2004). Sleep-dependent declarative memory was then tested using a two-dimensional visual spatial memory task which involved being trained to criterion and then given a cued recall test after a period of wake or overnight sleep. Both groups demonstrated more consolidation following sleep compared to wakefulness, indicating that sleep aids hippocampally mediated memory consolidation in children regardless of their neurodevelopment. Nevertheless, participants with ASD had poorer overall memory consolidation than TD participants. Specifically, children with ASD had impairments in spatial memory, poorer immediate recall and overall memory recall than TD participants. Importantly, better-quality sleep in the ASD group correlated with memory consolidation, showing a direct link between sleep disturbances and detrimental effects on cognitive functioning in people with ASD. Therefore, Maski et al. (2015) suggest that if sleep quality is increased in children with ASD, it could offer another way to help improve memory retention and cognitive functioning. The study received a good score on the SQAC, (19/28= 67.9%); however, the sample size was small (ASD  $n=22$ ) when considering the age range (9-16 years) and, although age-matched with the control group ( $n=20$ ), results should be treated with caution.

Limoges et al.'s (2013) study explored the association between sleep and cognitive daytime performance in young adults with ASD and TD. The results were akin to findings from the two aforementioned studies (Bruni et al., 2007; Maski et al., 2015); ASD participants had poorer sleep and more self-reported sleep disturbances compared to TD participants. In addition, the ASD group performed as accurately as the TD group in the memory and attention tasks; however, their performance was slower. Interestingly, despite having slower

response times, the ASD group made fewer mistakes than the TD group in tasks that involved selective attention and sensory-motor memory. The authors explained that these results could be the result of alternative strategies that people with ASD utilised to overcome the difficulties they would have using the strategies of TD people perhaps indicating that people with ASD take different neural pathways and have altered connectivity substrates in certain tasks. We scored the Limoges et al. (2013) study 20/28 (71.45%) on the SQAC, and it would have scored higher with a larger sample size ( $n=17$ ) of people with ASD and a clearer estimate of variance in the main results.

Finally, Taylor et al.'s (2012) study investigated sleep and cognitive functioning in 335 children with ASD or pervasive developmental disorder-not otherwise specified (PDD-NOS; a subtype of ASD prior to DSM-5). The aim of the study was to describe the associations between sleep and subsequent daytime cognitive and adaptive functioning. The study found that those with shorter sleep duration demonstrated impaired intelligence, perceptual and verbal skills, compared to children who slept longer. In children with ASD, sleep disturbances had a detrimental effect on the ability to learn, understand and communicate. Children with ASD who slept for fewer hours and suffered from (parent-reported) night-time breathing problems exhibited less aptitude to complete nonverbal tasks (e.g., puzzles, mazes, block building), highlighting a possible association between sleep apnea and perceptual tasks for children with ASD. Treatment for apnea could therefore improve cognitive functioning. Children with ASD who, on average, slept more per night without waking, demonstrated a better ability to learn, understand and communicate with others during the day. The Taylor et al.'s (2012) study received a score of 16/28 (57.1%) on the SQAC because the outcome measures were not well defined due to the numerous materials utilised. This could have led to

problems with controlling for confounding variables due to the number of tests and results from the various materials used, which may have led to a misclassification bias.

Overall, all the literature reviewed had good SQAC scores and used quantitative methods to investigate the effects of sleep disturbances on cognitive functioning in people with ASD. There was a lack of qualitative research which could have added exploratory depth to the review. In summary, from the literature reviewed, the cognitive functions that are specifically affected by a disruption of sleep in people with ASD comprise memory consolidation, declarative memory, selective attention, spatial memory, poorer immediate recall, overall memory recall, impaired intelligence and verbal skills.

### **What are the principal challenges associated with studying the impact of sleep disturbances in people with ASD?**

In order to answer this question, four main themes have been highlighted: ‘differences in psychometric measures’, ‘under-reporting sleep disturbances in people with ASD’, ‘the role of carers and family members’ and ‘barriers to research investigating sleep in people with ASD’.

#### **Theme 1: Differences in psychometric measures**

Five studies (Delahaye et al., 2014; Goldman et al., 2012; Hollway et al., 2013; Krakowiak et al., 2008; Maski et al., 2015) used the Children’s Sleep Habits Questionnaire (CSHQ) (Owens et al., 2000). The CSHQ is a validated measure with adequate internal consistency for a clinical sleep disorder sample ( $\alpha = .78$ ) and a community sample ( $\alpha = .68$ ), whereas test-retest reliability ranged from .62 to .79 (Owens et al., 2000). It consists of 45 items that relate to clinical sleep complaints exhibited over the most recent typical week, yielding scores for

bedtime resistance, sleep onset delay, sleep duration, night awakenings, sleep anxiety, daytime sleepiness, parasomnias, and sleep-disordered breathing.

Three studies (Polimeni et al., 2005; Schreck et al., 2004; Taylor et al., 2012) used the parent reported Behavioural Evaluation of Disorders of Sleep (BEDS) questionnaire (Schreck et al., 2003). The BEDS is a 28-item parent-report questionnaire on sleep quality, developed for use with children (5-12 years). Three questions address general characteristics of children's sleep, and one question asks whether the parent identifies their child as having poor sleep. This could be an interesting question to see how the parents interpret their child's sleep. Parents' answers on the BEDS relate to their child's sleep behaviours over the last six months. The BEDS has adequate internal consistency ( $\alpha = .82$ ) (Schreck et al., 2003) and identifies children with sleep disturbances and those without (Taylor et al., 2012).

Four of the reviewed studies (Bruni et al., 2007; Maski et al., 2015; Miano et al., 2007; Wiggs & Stores, 2004) used subjective measures of sleep in addition to PSG. These measures included a sleep questionnaire developed by the authors (Miano et al., 2007), Simonds and Parraga Sleep Questionnaire (Miano et al., 2007), Pediatric Daytime Sleepiness Scale (PDSS) (Bruni et al., 2007) and actigraphy (movement monitoring) (Maski et al., 2015). A sleep diary was also utilised as one of the subjective measures of sleep. Patzold et al. (1998) used a sleep diary in which various questions about the participant's sleep were included, while Quist et al. (2015) used a sleep diary together with the Pittsburgh Insomnia Rating Scale 20 (PIRS 20). The final two studies (Elia et al., 2000; Limoges et al., 2013) out of the 15 studies did not use any psychometric measures, using just PSG. More specifically, Elia et al. (2000) did not report the strength of correlations in their study, and therefore, the results must be regarded with caution.

In summary, all the studies reviewed in this section had very good quality scores (see Table 1); however, the variety of subjective and objective measures used in each study make it difficult to draw like-for-like comparisons between them. Although data from objective and subjective sleep measures have been found to be consistent, studies using only subjective measures such as sleep questionnaires or sleep diary can potentially suffer from biases due to parental reporting. Therefore, research would benefit from a greater use of objective measures such as actigraphy or PSG recordings, or a combination of both objective and subjective measures of sleep.

### **Theme 2: Under-reporting sleep disturbances in people with ASD**

Only three studies (Delahaye et al., 2014; Miano et al., 2007; Wiggs & Stores, 2004) indicated why sleep disturbances are often under-reported in people with ASD. According to Delahaye et al. (2014), these difficulties stem from the variability in ASD presentation, differences in participants' age range and the use of different ASD diagnostic criteria. Delahaye and colleagues noted that their findings were more likely to represent the experience of ASD children at autism clinics in academic medical centers, rather than the experience of all types of ASD individuals. Further, that they might have excluded a number of potential ASD children who lived in rural areas and/or those without health insurance. Similarly, Miano et al. (2007) excluded participants with ASD whose parents reported habitual snoring, respiratory sleep disturbances, abnormal sleep patterns, craniofacial abnormalities, obesity or respiratory sleep disorders from their study to improve standardisation of participants. However, this highlights that co-occurring sleep disorders may be under-represented in ASD studies and therefore, may not be an accurate reflection of the overall population of people with ASD. For this reason, we scored the Miano et al. study

18/28 (64.3%) on the SQAC. Although the study included an adequate sample size of 31 children with ASD with age-matched controls, results should be treated with caution.

Further, Wiggs and Stores (2004) state that the parents of children with ASD may be biased in their recording of sleep disturbances. This is suggested by the discrepancies between actigraphy data and parental reports. An actigraph is a small device that continuously monitors movement. An algorithm is used to determine sleep and wake, based on movement, to indicate the quality and quantity of sleep (Morgenthaler et al., 2007). Wiggs and Stores (2004) discussed the importance of making a distinction between general sleep problems which are often minor or transient, and clinical sleep disorders requiring treatment. This distinction would help indicate more appropriate clinical interventions. The study scored high on the SQAC (78.6%) due to its large sample size (ASD  $n=69$ ); however, it failed to elaborate on the age-matched control participants. The authors acknowledged that their study was limited by the subdivision of the sample to control for or explore other pertinent variables.

These studies indicate that sleep disturbances are often under-reported in people with ASD. This may be because those with ASD often have a wide range of presentations and sleep disturbances may result from an unaddressed problem in the child, family, or environment (Delahaye et al., 2014). Those with comorbid presentations may also be removed from sleep studies to make the sample more homogeneous (Delahaye et al., 2014; Miano et al., 2007). Wiggs and Stores (2004) indicated that parents might be unaware of the sleep disturbances their child may be experiencing and the use of objective measures of sleep may resolve this. All the aforementioned studies focused on children, and there is a gap in the literature for understanding why sleep disturbances are under-reported in adults with ASD.

### **Theme 3: Role of carers and family members**

Four of the reviewed studies investigated how carers/ family members were affected by the disrupted sleep of people with ASD (Krakowiak et al., 2008; Polimeni et al., 2005; Quist et al., 2015; Wiggs & Stores, 2004). For instance, Polimeni et al. (2005) suggest that behaviour analysis followed by behavioural interventions are more effective for young children with ASD. They discussed that parents of children with ASD may be experienced at using behavioural techniques with their child's problematic behaviours and may be more experienced in delivering behavioural interventions than other techniques. Polimeni et al. (2005) conclude that behavioural interventions may have better outcomes for sleep disturbances in children with ASD than other approaches such as Cognitive Behavioural Therapy. Quist et al. (2015) also found that their psychoeducation sleep-hygiene group for adults with a diagnosis of ASD helped improve sleep disturbances. However, we scored this study marginally on the SQAC (14/28=50%) as this was a small study with only 14 male participants with ASD, and it did not specify how the participants were educated or selected, estimates of variance in the main results or how the authors controlled for the confounding factors.

Wiggs and Stores (2004) suggest that discrepancies between actigraphy data and parental reports may be due to the parents' sleep also being disturbed, and that parents will only be aware of their child's sleeplessness if the child signals their awake state to the parent. They suggest that while mental states of mothers of children with ASD and mothers of children with TD did not significantly differ, it is possible that their disturbed sleep have an impact on their wellbeing. This could support the idea that sleep interventions focused on altering a child's behaviour or cognitions regarding sleeplessness might also be of benefit to the carers/family members with disrupted sleep. Wiggs and Stores (2004) also discussed how

children with ASD experienced sleep disturbances because of sensory difficulties including auditory or tactile sensitivities which may result in wrapping themselves tightly in a duvet or refusing to have covers touch their bodies. Wiggs and Stores suggest that parents may not be aware of these problems and therefore the lack of awareness means that the parents do not consider interventions such as using soft bedding or minimising noise. For example, children with ASD showed adverse reactions to textures that were not soft (Paron-Wildes, 2008) and have a preference for soft cotton against their skin rather than hard surfaces (Williams & Vouchila, 2003). Therefore, a cognitive and behavioural assessment may benefit those people with ASD who find themselves awake during the night, to understand patterns and how to break any maintaining factors. Wiggs and Stores (2004) conclude that further investigation of this phenomenon may result in developing novel sleep interventions.

Krakowiak et al. (2008) found that parents of children with TD rarely reported their child's sleep problems as affecting the family's daily functioning. However, a high proportion of parents of children with ASD (one in five families) did report problems with family functioning because of their child's sleep disturbances. The authors highlighted a lack of data concerning the impact that sleep disturbances on cognitive functioning have on families, carers, and the person with ASD, possibly resulting from a lack of qualitative research. This study is important as the mental health and well-being of the carers/parents is a critical factor when it comes to taking care of individuals with ASD. More knowledge into the (disrupted) family's daily functioning would provide more opportunities in developing interventions or behavioural strategies that would improve the quality of life and well-being of both the family and the affected individual (Halstead et al., 2021).

In conclusion, carers and other family members are affected by the disrupted sleep of people with ASD, and therefore sleep interventions aimed at children might also help educate carers

and family members on how to manage their sleep disturbances. The focus on children here highlights the lack of research into the impact of sleep disturbances on adults with ASD and their families and carers. Despite this limitation, all studies scored high on the SQAC. For example, Polimeni et al. (2015) study received a score of 19/28 (67.9%) and would have scored higher if the authors had included more information on participants selection, study design, estimates of variance in the main results, and control for confounding factors.

#### **Theme 4: Barriers to research investigating sleep in people with ASD**

Four of the 15 studies explicitly mentioned the risk and ethical implications of studying sleep disturbances in people with ASD (Delahaye et al., 2014; Krakowiak et al., 2008; Limoges et al., 2013; Wiggs & Stores, 2004). Delahaye et al. (2014) found that children with ASD who have more significant sleep problems are more likely to experience reduced health-related quality of life, as reported by their parents. The authors used the Health-Related Quality of Life (HRQoL) questionnaire which emphasises an individual's subjective perception of health and wellbeing rather than objective indicators of health, thus findings should be interpreted with caution.

Similar to Delahaye et al. (2014), Krakowiak et al. (2008) also discussed the limitations of relying on subjective parent reporting of their child's sleep disturbances. Krakowiak et al. (2008) suggest that while parent reports are correlated with objective actigraphy of sleep routines in children with ASD, this is the case only if they are made aware of the disturbed sleep by their child. Limoges et al. (2013) and Wiggs and Stores (2004) also highlighted that sleep disturbances in 'non-complaining' persons with ASD is under-reported, as the parents are often unaware. This suggests that individual consideration or a form of objective monitoring should be given to children with ASD who have limited communication abilities.

Further, Wiggs and Stores (2004) study benefited from using an objective sleep measure (actigraphy) to understand the nature of the sleep disturbances in people with ASD and further develop the knowledge around the mechanisms underlying successful sleep interventions. The authors found that children with ASD often have high levels of anxiety, challenging daytime behaviour, use of inappropriate or fixed routines and rituals, and impaired communication and social skills. All these factors could result in difficulties with the implementation of specific behavioural sleep interventions and a possible reduction in their efficacy.

In summary, these studies suggest that research would benefit from using objective measures of sleep in individuals with ASD, since parent report may not provide an accurate representation of sleep difficulties. Where studies rely only on subjective parent report sleep measures the results could be misinterpreted and provide misleading evidence of sleep disturbances in people with ASD (Wiggs & Stores, 2004). These studies also had very good SQAC scores. For example, Krakowiak et al. (2008) received a high score of 22/28 (78.6%), and would have scored higher if the authors had reported some estimates of variance in the results. The Delahaye et al. (2014) study was also scored quite well (20/28= 71.4%) as the sample size consisted of 86 parents of children with ASD.

## **Discussion**

### **Significance of the Main Findings**

This review aimed to identify what cognitive functions are impacted upon by sleep disturbances in people with ASD, the principal challenges associated with studying the impact of sleep in people with ASD, and critically review the quality of the research. These

findings highlight the adverse effects of sleep disturbances on daytime behaviour, cognitive and general functioning in people with ASD as well as the negative impact on their families/carers. Sleep disturbances in people with ASD has a detrimental effect on different cognitive functions such as memory consolidation, declarative memory, selective attention, spatial memory, poorer immediate recall, overall memory recall, impaired intelligence and verbal skills (Bruni et al., 2007; Limoges et al., 2013; Maski et al., 2015; Taylor et al., 2012). Importantly, when sleep improves, it has a positive effect on cognitive functioning (Limoges et al., 2013; Taylor et al., 2012). Also highlighted was the different neural pathways that people with ASD have when comparing cognitive functioning in people with TD. Therefore this review has highlighted a possible methodological problem when comparing the cognitive functioning between those with ASD and those with TD. A clearer understanding of these associations may help develop measures and interventions that may help with these areas.

The literature review also highlighted the problems of identifying sleep disturbances in people with ASD. As there is such variation in ASD presentations including a wide range of communication difficulties, this variation may result in sleep disturbances being under-reported by people with ASD (Delahaye et al., 2014; Wiggs & Stores, 2004). Co-occurring conditions alongside the ASD diagnosis also complicate this under-reporting. Many people with ASD are often excluded from studies investigating sleep disturbances which may result in their under-representation in the broader literature. Therefore, these complex presentations should be considered and represented in research assessing sleep or developing measures of assessment or treatment.

Further, this review highlighted a number of measures that researchers use to investigate sleep problems. Different psychometric measures that are used to identify sleep disturbance in children with ASD include subjective questionnaires, sleep diaries, actigraphy and PSG

recordings. Moore et al. (2017) emphasised the importance of using an objective measure as well as a subjective measure such as parental reports to screen for sleep disturbances in children with ASD since the parental recording alone may not be accurate. Despite increased costs associated with using multiple measures, with advances in technology actigraphy and PSG recordings may become less expensive and more user-friendly and may help with accurately diagnosing sleep disorders in the ASD population. Schreck and Mulick (2000) found that parents of children with ASD reported significantly more sleep problems in their child, which is different to the perceptions of parents from other study groups (e.g. children with General Mental Retardation, and children with TD). Exploration in this area could also help provide more rich data regarding how parents and carers currently deal with sleep disturbances in those with ASD. More research also needs to focus upon appropriate measures of sleep in people with ASD that take into consideration the impact on cognitive functioning rather than just focusing the measure on behaviours. The area is currently under-researched and therefore clinicians, parents and carers may not be aware of the impact of sleep disturbances on cognitive functioning.

### **Summary of methodological constraints and research limitations**

This review found that, in people with ASD, the effects of sleep disturbances on cognitive functioning are complex and require further investigation. Data are limited regarding the impact that impaired cognitive functioning due to sleep disturbances on families, carers and the person with ASD. Not one of the reviewed studies employed a qualitative research design. A qualitative approach involving interviews with these parties could explore the impact that impaired cognitive functioning, due to sleep disturbances, on daily functioning

and wellbeing. This qualitative approach may also reveal successful parental approaches in improving sleep quality in children with ASD.

Another limitation is that several studies did not report the IQ or severity of symptoms of the participants in their studies entirely (Delahaye et al., 2014; Wiggs & Stores, 2004). This is important as several studies have highlighted IQ and symptom severity in children with pervasive developmental disorders as the main predisposing factors for the development of insomnia (Cohen et al., 2014; Hollway et al., 2013). This missing information means that the possible links between IQ and sleep disturbances could not be established.

Finally the use of the key terms could have limited this review. There are a lot of different terms for sleep disturbances, ASD and cognitive functioning. This might have caused the search of the literature to be too narrow. A possible way to overcome this limitation in the future is to be more specific in searching for literature – for example, look at physical problems causing sleep disturbances e.g., apnea. Also, key terms for cognitive functioning could be more specific e.g., attention, working memory, and finally key terms for sleep could include insomnia, night terrors etc.

### **Clinical implications and future research recommendations**

This review has highlighted that, for accurate reporting of sleep disturbances in children with ASD, both objective and subjective measures of sleep are recommended as they can counter misinterpretation and provide additional insight into the nature of the sleep disturbances (Krakowiak et al., 2008; Wiggs & Stores, 2004). This review has also highlighted that there is a gap in the literature regarding the impact that sleep disturbances in children with ASD have on their family/carers' wellbeing. Therefore, additional measures regarding

family/carers' wellbeing and stress would help to identify any risk factors that lead to a proliferation of parenting stress (Benson, 2006). The use of a wide range of different neuropsychological tests would provide a more in-depth view of the impact of sleep disturbances on cognitive functioning rather than just focusing on behavioural dysfunction. This multidimensional approach to sleep dysfunction in people with ASD will help identify risk factors and provide more accurate data. This, in turn, might help identify the most appropriate sleep intervention for the person with ASD. Further, this literature review has found a gap in the literature for the most appropriate clinical interventions for this client group. Future research might compare sleep interventions to identify which are the most effective for this group.

## **Conclusion**

This literature review has highlighted that cognitive functions in people with ASD are negatively affected by sleep disturbances. Reduced cognitive functioning in people with ASD has a major impact on their daily functioning, as well as impacting upon the families and carers. There are a limited number of standardised measures for identifying sleep disturbances in this population, alongside methodological problems relying on subjective parental reports; thus, we recommend the use of objective sleep measures. Finally, there are a lack of evidence-based clinical interventions to help improve sleep disturbances in people with ASD, along with a lack of parental accounts regarding successful methods of being able to manage the sleep disturbances.

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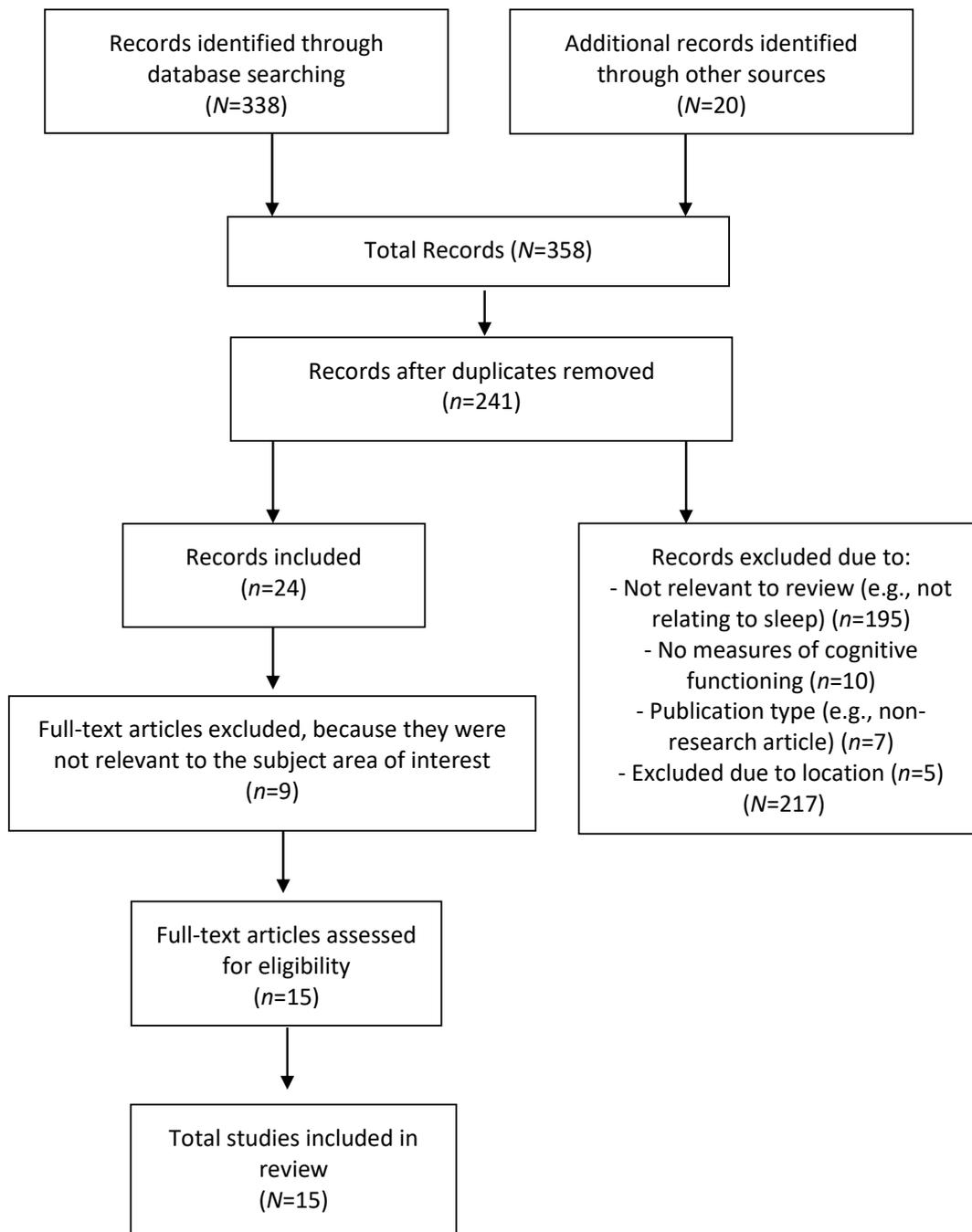
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Tables and Figures.



*Figure 1.* Preferred Reporting Items for Systematic Reviews and Meta-analyses (PRISMA)

flow diagram illustrating the study selection process.

Table 1

*Summary of the SQAC scores for the reviewed papers*

<b>Author</b>	<b>Date</b>	<b>Inter-rater reliability (Kappa statistic)</b>	<b>Standard Quality Assessment Criteria (SQAC) Scores</b>
Bruni, O., Ferri, R., Vittori, E., Novelli, L., Vignati, M., et al	2007	.682  $p < .001$	22/28= 78.6%
Delahaye, J., Kovacs, E., Sikora, D., Hall, T., Orlich, F., et al	2014		20/28= 71.4%
Hollway, J., Aman, M., Butter, E.	2013	.682  $p < .001$	22/28= 78.6%
Limoges, É., Bolduc, C., Berthiaume, C., Mottron, L., Godbout, R.	2013		20/28= 71.45%
Maski, K., Holbrook, H., Manoach, D., Hanson, E., Kapur, K., et al	2015	.714  $p < .001$	19/28= 67.9%
Miano, S., Bruni, O., Elia, M., Trovato, A., Smerieri, A., et al	2007		18/28= 64.3%
Patzold, L., Richdale, A., Tonge, B.	1998	.868  $p < .001$	18/28= 64.3%
Quist, H., Chaplin, E., Hendey, O.	2015		14/28 = 50%

Taylor, M., Schreck, K., Mulick, J.	2012	.75 <i>p</i> < .001	16/28= 57.1%
Elia, M., Ferri, R., Musumeci, S., Del Gracco, S., Bottitta, M., et al	2000		19/28= 67.9%
Goldman, S., Richdale, A., Clemons, T., Malow, B.	2012	.509 <i>p</i> < .009	19/28= 67.9%
Krakowiak, P., Goodlin-Jones, B., Hertz-Picciotto, I., Croen, L., Hansen, R.	2008		22/28= 78.6%
Polimeni, M., Richdale, A., Francis, A.	2005	1.000 <i>p</i> < .001	19/28= 67.9%
Schreck, K., Mulick, J., Smith, A.	2014	.559 <i>p</i> < .003	17/28= 60.7%
Wiggs, L., Stores, G.	2004		22/28= 78.6%

## Supplementary material

Table 1

*Characteristics of reviewed studies*

<b>Author/Date/ Location/ Quality Assessment Rating</b>	<b>Study Aim</b>	<b>Research Design/Sampling Method</b>	<b>Sample population</b>	<b>Method of data collection/ data analysis</b>	<b>Key Findings</b>
Bruni,O., Ferri,R., Vittori,E., Novelli,L., Vignati, M., et al.  2007	To analyse sleep in children with AS and Autism	Cross-sectional study  Opportunity Sampling	8 AS (7 male, 1 female; Age (years) <i>M</i> =12.7, <i>SD</i> =2.6 years, <i>range</i> =7-15)  10 Autistic (9 male, 1 female; Age (years)	Sleep Questionnaire, PDSS, ADOS, CBCL, WISC- III, PSG  Mann-Whitney test, Cohen's & Spearman correlation	Children with AS show peculiar sleep patterns compared to control. Short sleep duration (<9 h) was almost twofold (59% vs 32%), and the risk for sleep onset problems more than fivefold (53% vs 10%) more common in the AS group than in the control group.  AS Vs Controls $U=0.023$ $p<0.05$ , $d=1.35$ AS Vs Autism $U=0.041$ $p<0.05$ , $d=1.24$

<p>Italy</p> <p>QR=78.6% (Kappa=0.68)</p>			<p><math>M=11.9</math>, <math>SD=2.5</math> years, <i>range</i>=7-15)</p> <p>12 Control matched for age (5 female, 7 male, Age (years) <math>M=12.6</math>, <math>SD=3.7</math> years, <i>range</i>=7-15)</p>		<p>Stages of sleep were significantly different between Autism, Asperger and controls.</p> <p>The following significant associations between PSG &amp; Behavioural/Cognitive measures were found:</p> <ul style="list-style-type: none"> <li>-REM latency &amp; Full-scale IQ, <math>r_s=89</math>, <math>p&lt;0.05</math></li> <li>-REM latency &amp; Performance IQ, <math>r_s=0.86</math>, <math>p&lt;0.05</math></li> <li>-REM % &amp; CBCL Internalising, <math>r_s=0.81</math>, <math>p&lt;0.05</math></li> <li>- SE% &amp; CBCL Externalising, <math>r_s=0.95</math>, <math>p&lt;0.05</math></li> </ul>
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<p>Delahaye, J., Kovacs, E., Sikora, D., Hall, T., Orlich, F., et al.</p> <p>2014</p> <p>USA</p> <p>QR=71.4%</p>	<p>To investigate the associations between Health-related quality of life (HRQoL) and overall sleep problems within the context of critical clinical characteristics in children with ASD</p>	<p>Cross-sectional study</p> <p>Opportunity Sampling</p>	<p>86 parents of children (72 male, 14 female; Age (years) <i>Mean</i>=7.18, <i>range</i>=4-12)</p> <p>4 AS</p> <p>57 Autistic</p> <p>21 PDD-NOS</p> <p>4 participants dropped out</p>	<p>PedsQL, CSHQ, CBCL, PDDBI, SB5 &amp; MSEL</p> <p>T-tests, Pearson correlations, linear regression models</p>	<p>Correlations were found between the sleep duration scale and the PedsQL total <math>r=-0.36</math>, <math>p&lt;.001</math> and psychosocial summary <math>r=-0.36</math>, <math>p&lt;.001</math> scores.</p> <p>Sleep duration/anxiety was significantly predicted by:</p> <ul style="list-style-type: none"> <li>- PedsQL physical summary score (<math>\beta=-0.73</math>, <math>p&lt;0.01</math>)</li> <li>- PedsQL total score (<math>\beta=-1.61</math>, <math>p&lt;0.05</math>)</li> <li>- CHSQ total (<math>\beta=-2.16</math>, <math>p&lt;0.01</math>)</li> <li>- CHSQ psychosocial (<math>\beta=-2.11</math>, <math>p&lt;0.01</math>)</li> </ul>
<p>Hollway, J., Aman, M., Butter, E.</p> <p>2013</p>	<p>To investigate if:</p> <ul style="list-style-type: none"> <li>- an inverse association existed between disturbed sleep, intellectual functioning,</li> </ul>	<p>Cross-sectional study</p> <p>Opportunity Sampling</p>	<p>1,583 children (1327 male, 256 female; Age (years) <i>M</i>=6.34,</p>	<p>CSHQ, VABS, MSEL, SB5, CBCL, ADOS &amp; SSP</p>	<p>Anxiety, autism symptom severity, sensory sensitivities, and GI problems were associated with sleep disturbance, <math>F(5,1430)=33.101</math>, <math>p&lt;.001</math>.</p> <p>IQ positively predicted sleep disturbance, and children with Asperger's Disorder (AD)</p>

<p>USA</p> <p>QR=78.6% (Kappa=0.68)</p>	<p>adaptive behaviour, age, and parent education</p> <p>- a positive association existed between sleep disturbance, autism symptom severity, and internalising and externalising behaviour</p> <p>- medical issues such as epilepsy, GI problems, and medication use, were positively associated with sleep disturbance</p>		<p><math>SD=3.5</math>, <math>range=2-17</math>).</p> <p>152 AD</p> <p>1032 Autistic</p> <p>399 PDD-NOS</p>	<p>T-tests, ANOVA, Hierarchical Regression Models</p>	<p>were more vulnerable than others <math>R^2=0.193</math>; <math>F(12, 1376)=27.345, p&lt;0.001</math></p>
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<p>Limoges, É., Bolduc, C., Berthiaume, C., Mottron, L., Godbout, R.</p> <p>2013</p> <p>Canada</p> <p>QR=71.5%</p>	<p>To investigate if markers of poor sleep, documented in ASD, correlate with non-verbal cognitive performance</p>	<p>Randomised Control Trial</p> <p>Opportunity Sampling</p>	<p>17 ASD (9 with high-functioning autism and 8 with AS (16 male, 1 female; Age (years) <math>M=21.7</math>, <math>SD=3.5</math>, <math>range=16-27</math>)</p> <p>14 Controls (13 male, 1 female; Age (years) <math>M=21.8</math>, <math>SD=4.1</math>, <math>range=16-27</math>)</p>	<p>Sleep questionnaire, WAIS-III, BDI-II, STAI, AYSRS, FCRTT, CblTa, FLS-BEM144, PRPT &amp; TLT</p> <p>Spearman's correlation</p>	<p>Significant correlations were found for controls between: Selective attention</p> <ul style="list-style-type: none"> <li>- TR total and sleep <math>r_s=-0.82</math>, <math>p&lt;0.01</math></li> <li>- Errors and sleep <math>r_s=-0.59</math>, <math>p&lt;0.05</math></li> </ul> <p>Declarative episodic memory and sleep</p> <ul style="list-style-type: none"> <li>- Recall 1 and %SWS, <math>r_s=0.60</math>, <math>p&lt;0.05</math></li> <li>- Recall 2 and %SWS, <math>r_s=0.65</math>, <math>p&lt;0.05</math></li> <li>- Recall 3 and %WASO, <math>r_s=-0.55</math>, <math>p&lt;0.05</math>; and %SWS, <math>r_s=0.61</math>, <math>p&lt;0.05</math></li> <li>- delayed recall and %WASO, <math>r_s=-0.53</math>, <math>p&lt;0.05</math></li> </ul> <p>Sensory-motor and cognitive procedural memory and sleep</p> <ul style="list-style-type: none"> <li>- learning phase and %SWS 3<sup>rd</sup>, <math>r_s=-0.75</math>, <math>p&lt;0.01</math></li> </ul>
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					<p>Significant correlations were found for ASD between:</p> <ul style="list-style-type: none"> <li>- Declarative episodic memory Recall 1 and: <ul style="list-style-type: none"> <li>- Sleep latency, <math>r_s=-0.62</math>, <math>p&lt;0.01</math></li> <li>- % stage 1, <math>r_s=-0.47</math>, <math>p&lt;0.05</math></li> <li>- % WASO, <math>r_s=-0.54</math>, <math>p&lt;0.05</math></li> </ul> </li> <li>- learning phase and: <ul style="list-style-type: none"> <li>- %SWS 3<sup>rd</sup>, <math>r_s=-0.53</math>, <math>p&lt;0.05</math></li> <li>- C3SS, <math>r_s=-0.66</math>, <math>p&lt;0.01</math></li> </ul> </li> <li>- Selective attention <ul style="list-style-type: none"> <li>- RT total and SL, <math>r_s=0.71</math>, <math>p&lt;0.01</math></li> <li>- Errors and SL, <math>r_s=0.55</math>, <math>p&lt;0.05</math>; and %S1, <math>r_s=0.59</math>, <math>p&lt;0.05</math></li> </ul> </li> <li>- Memory <ul style="list-style-type: none"> <li>- Initial time and %S1, <math>r_s=-0.61</math>, <math>p&lt;0.05</math></li> <li>- Execution time and SL, <math>r_s=0.62</math>, <math>p&lt;0.05</math></li> <li>- % correct and SL, <math>r_s=-0.55</math>, <math>p&lt;0.05</math></li> </ul> </li> </ul>
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<p>Maski, K., Holbrook, H., Manoach, D., Hanson, E., Kapur, K., et al</p> <p>2015</p> <p>USA</p> <p>QR=67.9% (Kappa=0.71)</p>	<p>To examine the role of sleep in the consolidation of declarative memory in children with ASD</p>	<p>Case-control study</p> <p>Opportunity Sampling</p>	<p>22 ASD (19 male, 3 female; Age (years) <math>M=11.3</math>, <math>SD=2.1</math>, <math>range=9-16</math>)</p> <p>20 TD (18 male, 2 female; Age (years) <math>M=12.3</math>, <math>SD=2.1</math>)</p>	<p>Actigraph, PSG, VSMT, CBCL, SRS, DAS-II, NVIQ, D- KEFS, CSHQ &amp; IFMS</p> <p>Unpaired t- tests, Wilcoxon rank sum test, Fisher exact test, mixed- effects regression, Pearson correlation tests</p>	<p>Memory performance deteriorated significantly more across the Wake condition than in the Sleep [Wake: 16-25.8, Sleep:0.7- 8.7, <math>F(1,40)=7.95</math>, <math>p&lt;0.01</math>] and the ASD group demonstrated poorer overall memory recall than participants with TD [ASD: 16.7- 26.9, TD:2.2-8.6, <math>F(1,40)=6.2</math>, <math>p&lt;0.02</math>]</p>
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<p>Miano, S., Bruni, O., Elia, M., Trovato, A., Smerieri, A., et al</p> <p>2007</p> <p>Italy</p> <p>QR=64.3%</p>	<p>To evaluate sleep in children with ASD with sleep questionnaires and PSG</p>	<p>Randomised Control Trial</p> <p>Opportunity Sampling</p>	<p>31 ASD (28 male, 3 female; Age (years) <i>M</i>=9.53, <i>SD</i>=3.82, <i>range</i>=7–19 years)</p> <p>18 age- matched controls (9 male, 9 female; Age (years) <i>M</i>= 10.2 years, <i>SD</i>=2.93, <i>range</i>=7–19 years)</p>	<p>PSG, WISCR, WAIS-III &amp; Sleep questionnaire</p> <p>Pearson's chi- squared, Mann– Whitney</p>	<p>Compared to typical development in children the ASD children were significantly different on:</p> <ul style="list-style-type: none"> <li>- Sleep duration &lt; 8 hrs, <math>X^2=5.55</math>, <math>p&lt;0.02</math></li> <li>- Latency to sleep, <math>X^2=16.42</math>, <math>p&lt;0.00001</math></li> <li>- Difficulty getting to sleep, <math>X^2=9.87</math>, <math>p&lt;0.002</math></li> <li>- Drinks stimulant beverages in the evening, <math>X^2=63.12</math>, <math>p&lt;0.00001</math></li> <li>- Fluids or drugs to facilitate sleep, <math>X^2=81.58</math>, <math>p&lt;0.00001</math></li> <li>- Hypnic jerks, <math>X^2=48.78</math>, <math>p&lt;0.00001</math></li> <li>- Poor sleep quality, <math>X^2=117.46</math>, <math>p&lt;0.00001</math></li> <li>- More than two awakenings per night, <math>X^2=3.91</math>, <math>p&lt;0.05</math></li> <li>- Waking up to drink or eat in the night, <math>X^2=5.94</math>, <math>p&lt;0.015</math></li> </ul>
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					<ul style="list-style-type: none"><li>- Difficulty to fall asleep after awakenings, <math>X^2=25.31, p&lt;0.00001</math></li><li>- Bedwetting, <math>X^2=41.72, p&lt;0.00001</math></li><li>- Daytime somnolence, <math>X^2=4.69, p&lt;0.03</math></li><li>- Falling asleep at school, <math>X^2=5.80, p&lt;0.02</math></li></ul> <p>Subtle alterations of NREM sleep was detected between groups (significance at <math>p&lt;0.05</math>):</p> <p>TIB: <math>u=0.044</math></p> <p>SPT: <math>u=0.014</math></p> <p>TST: <math>u=0.007</math></p> <p>RL: <math>u=0.02</math></p>
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<p>Patzold, L., Richdale, A., Tonge, B.</p> <p>1998</p> <p>Australia</p> <p>QR=64.3% (Kappa=0.87)</p>	<p>To investigate sleep problems in children with autism and explore the association between sleep problems and daytime behaviour.</p>	<p>Correlational design</p> <p>Opportunity Sampling</p>	<p>31 ASD (Autistic 25 male, 6 female; AS 6 male, 1 female; Age (months) <math>M=93.5</math>, <math>SD=31.5</math>, <math>range=44-152</math>)</p> <p>36 PDD (29 male, 7 female; Age (months) <math>Mean=101.2</math>, <math>SD=31.0</math>, <math>range=63-171</math>)</p>	<p>Sleep diary/questionnaire, DBC &amp; CBCL</p> <p>Pearson and point-biserial correlation</p>	<p>The type of past (<math>r=0.42</math>, <math>p&lt;0.01</math>) and present (<math>r=0.42</math>, <math>p&lt;0.01</math>) sleep problems reported by parents of children included night waking difficulties (<math>r=0.49</math>, <math>p&lt;0.01</math>), co-sleeping with parents, (<math>r=0.40</math>, <math>p&lt;0.01</math>), and poorer sleep quality (<math>r=0.45</math>, <math>p&lt;0.01</math>)</p>
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Quist, H., Chaplin, E., Hendey, O.  2015  UK  QR=50%	To determine the effectiveness of a psychoeducation sleep-hygiene group intervention for adults diagnosed with ASC, without a concurrent learning disability	Experimental Design  Opportunity Sampling	14 ASC (14 male, 0 female; Age (years) $M=28.16$ , $range=20-55$ )	PIRS 20  t-test	Significant improvements were seen on the PIRS-20 $t^{(*)}=2.51$ , $p<0.05$ and on follow-up measures of self-reported sleep distress $t^{(*)}=3.06$ , $p<0.05$
Taylor, M., Schreck, K., Mulick, J.  2012  USA	To investigate the associations between sleep behaviour in children with ASD and subsequent day-time cognitive and adaptive performance	Randomised control trial  Opportunity Sampling	335 PDD (296 male, 39 female; Age (years) $M=5.15$ , $SD=3.27$ , $range=1-18$ )	WPPSI, Leiter-R, DP-II, MSEL, SB5, SIB-R, VABS & BEDS  Pearson correlation, Stepwise	-Parental report of more hours slept per night singularly predicted better daily living skills ( $R^2=0.09$ ; $p<0.01$ ) and the combination of more total hours slept per night and hours napped during the day predicted better Adaptive Behaviour Composite ( $R^2=0.18$ ; $p<0.01$ ); motor skills ( $R^2=0.16$ ; $p<0.01$ ), and socialization ( $R^2=0.17$ , $p=0.01$ )

QR=57.1%			219 Autistic 116 PDD-NOS	multiple linear regression	-More hours slept per night in combination with fewer episodes of night waking with screaming and more sensitivity to sleeping environment disturbances significantly predicted children's higher communication scores ( $R^2=0.14$ , $p<0.01$ )
Elia, M., Ferri, R., Musumeci, S., Del Gracco, S., Bottitta, M., et al  2000  Italy  QR=67.9%	Evaluate the possible correlation between neurophysiological and psychological data in children with AD	Randomised control trial  *	17 male children with AD (Age (years) $M=10.36$ , $SD=3.79$ , $range=5$ years and 7 months-16 years and 8 months)  5 Control male subjects (Age (years) $M=9.22$ ,	Anamnesis, clinical examination, karyotyping, neurometabolic screenings, brain CT, MRI, PEP-R, CARS & PSG  Correlations	A negative* correlation between scores of passing items of PEP-R perception and SL ( $p<0.02$ ), Ssh ( $p<0.015$ ), FRL ( $p<0.03$ ), and %WASO ( $p<0.015$ ) was found together with a positive* correlation between the same items and TIB value ( $p<0.04$ ).  PEP-R eye-hand coordination passing items were correlated negatively* with Ssh ( $p<0.015$ ), FRL ( $p<0.02$ ), and %WASO ( $p<0.02$ ), and positively* with SPT ( $p<0.03$ ).

			<p><i>SD</i>=2.02, <i>range</i>=7.17-11.58)</p> <p>7 male subjects with fragile X syndrome and mental retardation (Age (years) <i>M</i>=9.92, <i>SD</i>=1.67, <i>range</i>=8.25-12 years)</p>		<p>CARS visual response was correlated negatively* with SPT (<math>p&lt;0.003</math>) and positively* with %WASO (<math>p&lt;0.03</math>); CARS non-verbal communication was negatively* correlated with TST (<math>p&lt;0.01</math>).</p> <p>People and activity level CARS items were correlated negatively* with REMd (<math>p&lt;0.01</math> and <math>p&lt;0.02</math>, respectively).</p>
Goldman, S., Richdale, A., Clemons, T., Malow, B.  2012	To identify areas defined by the parental report to be problematic in children with ASD	Cross-sectional study  Opportunity sampling	1859 ASD children (1571 male, 288 female; Age (Months) <i>M</i> =80.1,	ADOS, CSHQ & PCQ	Areas reported to be problematic include:  - Bedtime resistance  - Sleep onset delay  - Sleep duration  - Sleep anxiety

<p>USA</p> <p>QR=67.9% (Kappa=0.51)</p>			<p><i>SD</i>=42.3, <i>range</i>=36-216)</p> <p>- 67.2% Autistic</p> <p>- 9.8% AS</p> <p>- 23% PDD- NOS</p>		<ul style="list-style-type: none"> <li>- Night waking's</li> <li>- Parasomnias</li> <li>- Sleep-disordered breathing</li> <li>- Daytime sleepiness</li> </ul>
<p>Krakowiak, P., Goodlin-Jones, B., Hertz- Picciotto, I., Croen, L., Hansen, R.</p> <p>2008</p>	<p>To compare parent- reported sleep characteristics in young children with ASD aged 2 to 5 years to age- matched TD children as well as children with other developmental delays</p>	<p>Randomised control trial</p> <p>Opportunity sampling</p>	<p>163 TD (134 male, 29 female; Age (years) <i>M</i>=3.3)</p> <p>303 ASD (267 male, 36 female; Age (years) <i>M</i>=3.7)</p> <p>63 DD (46 male, 17</p>	<p>ADI-R, ADOS, MSEL, VABS, SCQ, CSHQ &amp; EEQ, Sleep questionnaire/h istory</p> <p>Multivariate Linear</p>	<ul style="list-style-type: none"> <li>- Children with ASD had higher sleep onset factor scores compared with TD children (<math>\beta=0.177, SE=0.091, p=0.05</math>)</li> <li>- Night waking factor scores were significantly elevated in the ASD group compared to TD children (<math>\beta=0.294, SE=0.090, p=0.001</math>)</li> </ul>

USA  QR=78.6%			female; Age (years) $M=3.6$	Regression Analysis	- Parent report of average sleep duration in a 24-hour period that children with ASD tended to sleep about half an hour less than TD children ( $\beta=-0.473$ , $SE=0.156$ , $p=0.003$ )
Polimeni, M., Richdale, A., Francis, A.  2005  Australia  QR=67.9% ( $Kappa=1.00$ )	- to compare sleep patterns in children with AS, autism, and TD  - to explore treatments used for sleep problems and examine treatment outcomes in these groups	Correlational design  Opportunity Sampling	66 TD (39 male, 27 female; Age (years) $M=6.0$ , $SD=3.1$ , $range=2-11$ )  52 AS (42 male and 8 female; Age (years) $M=9.3$ , $SD=3.1$ , $range=4-17$ )  53 Autistic (41 male and 12 female; Age (years) $M=6.5$ ,	Sleep questionnaire & BEDS  ANOVA & Tukey-Kramer post hoc tests	- TD group reported significantly fewer sleep problems than the other two groups [ $\chi^2(N=171)=9.618$ , $p=0.018$ ]  - There was a significant difference between the three groups on the disoriented waking factor of the BEDS ( $F(2,77)=4.145$ , $p=0.020$ ). Post hoc tests revealed the Asperger group had significantly higher scores on this factor than the TD group ( $M=8.07$ ( $SD=4.40$ ) vs $M=4.47$ ( $SD=3.69$ ), $p<0.05$ )  - There was a significant difference between the groups on the BEDS total score ( $F(2,$

			<p><i>SD=2.7, range=2-16)</i></p>	<p>81)=6.305, <math>p=0.003</math>). Post hoc tests indicated the Asperger group had significantly higher BEDS total scores than both the TD and the autism groups (<math>M = 116.85</math> (<math>SD=51.75</math>) vs <math>M = 73.48</math> (<math>SD=30.11</math>) vs <math>M = 79.53</math> (<math>SD=38.78</math>), <math>p&lt;0.01</math>)</p> <p>- Four categories were reported to be used for sleep problems: Behavioural intervention, medication, herbal treatment, and a mixed group of other treatments</p> <p>- Behavioural interventions were reported to be significantly more successful in the autism group than in the Asperger group (<math>F(2,32)=3.978</math>, <math>p=0.029</math>). Medication was rated as more successful by parents in the autism group and the Asperger group than by</p>
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					parents in the TD group ( $F(2, 39)=5.045$ ), $p=0.011$ )
Schreck, K., Mulick, J., Smith, A.  2004  USA  QR=60.7% ( $Kappa=0.559$ )	To provide a preliminary examination of whether specific sleep problems may be related to the expression of cardinal behavioural features of autism.	Correlational design  Opportunity Sampling	55 children with Autism (Age (years) $M=8:2$ , $SD=2:1$ , $range=5-12$ ) *Gender missing)	Sleep questionnaire, GARS & BEDS  Pearson correlations,  Stepwise multiple linear regression	- Communication problems were significantly related to increased sensitivity to stimuli in the sleeping environment and by periods of screaming during the night ( $R^2=0.18$ , $p<0.01$ )  - Fewer number of hours slept per night also predicted difficulties with social interactions ( $R^2=0.12$ , $p<0.01$ ) and overall diagnostic characteristics of autism ( $R^2=0.11$ , $p<:02$ )

Wiggs, L., Stores, G.  2004  UK  QR=78.6%	To investigate sleep disturbance in children with ASD	Exploratory Design  Opportunity sampling	69 ASD children (55 male, 14 female; Age <i>M</i> =9 years 4 months, <i>SD</i> =2 years 7 months, <i>range</i> =5-16)	Sleep history  Sleep diary  Actigraph  Child/Parental functioning questionnaire, SPSQ  Mann– Whitney U tests, T-test, Chi-squared test	- Behavioural sleep problems were more common in the younger age group than in the older children ( $\chi^2(1)=6.48, p=0.01$ )
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Autistic Disorder (AD), Autism Diagnostic Interview-Revised (ADI-R), Autism Diagnostic Observation Schedule (ADOS), Analysis of Variance (ANOVA), Autism Spectrum Condition (ASC), Autism Spectrum Disorder (ASD), Achenbach Youth Self-Report Scale (AYSRS), Beck Depression Inventory, 2nd edition (BDI-II), Behavioural evaluation of disorders of sleep (BEDS), Childhood Autism Rating Scale (CARS), Achenbach Child Behaviour Checklist (CBCL), Corsi block-tapping (CbITa), Children's Sleep Habits Questionnaire (CSHQ), Brain

Computed tomography (CT), Differential Abilities Scale II (DAS-II), Developmental Behaviour Checklist (DBC), Developmental Delays (DD), Tower and Trail Making subtests of the Delis-Kaplan Executive Function System (D-KEFS), Developmental Profile II (DP-II), Environmental Exposure Questionnaire (EEQ), Four choices reaction time test (FCRTT), Figure-learning subtest of the BEM 144 battery (FLS-BEM144), Gilliam Autism Rating Scale (GARS), Interest/Fatigue/Mood Scales (IFMS), Leiter International Performance Scale-Revised (Leiter-R), Magnetic resonance imaging (MRI), Mullen Scales of Early Learning (MSEL), Nonverbal IQ scores (NVIQ), Parental Concerns Questionnaire (PCQ), Pervasive Developmental Disorder (PDD), Pervasive Developmental Disorder Behaviour Inventory (PDDBI), Pervasive Developmental Disorder-Not Otherwise Specified (PDD-NOS), Paediatric Daytime Sleepiness Scale (PDSS), Paediatric Quality of Life Inventory 4.0 (PedsQL), Psychoeducational Profile Revised (PEP-R), Pittsburgh Insomnia Rating Scale 20 (PIRS 20), Photoelectric Rotary Pursuit Task (PRPT), Polysomnographic (PSG), Stanford-Binet Intelligence Scales, Fifth Edition (SB5), Social Communication Questionnaire (SCQ), Scales of independent behavior-revised (SIB-R), Simonds and Parraga Sleep Questionnaire (SPSQ), Social Responsiveness Scale (SRS), Short Sensory Profile (SSP), State-Trait Anxiety Inventory (STAI), Typical Development (TD), Tower of London task (TLT), Vineland adaptive behaviour scales (VABS), Visual spatial memory task (VSMT), Wechsler Adult Intelligence Scale – Third Edition Revised (WAIS-III), Wechsler Intelligence Scale for Children—Third Edition Revised (WISC-III), Wechsler Intelligence Scale for Children—Fourth Edition Revised (WISC-IV), Wechsler Intelligence Scale for Children Revised (WISCR), Wechsler Preschool and Primary Scale of Intelligence (WPPSI).

\* Missing data.

