

Sleep problems in autism spectrum disorders

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**SLEEP PROBLEMS IN AUTISM SPECTRUM DISORDERS: A COMPARISON TO
SLEEP IN TYPICALLY DEVELOPING CHILDREN USING ACTIGRAPHY,
DIARIES AND QUESTIONNAIRES**

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Abstract

It has been reported widely that children with Autism Spectrum Disorders (ASD) are more likely to experience sleep problems than children without ASD. The sleep of sixteen children with ASD, performance IQ in the normal range and a parentally-reported sleep problem was compared to a matched group of children without ASD. Five to seven nights of actigraphy data were collected for both groups, alongside sleep diaries and questionnaires. No group differences were identified through actigraphy or diary measures. Questionnaire data confirmed that the children with ASD had a higher prevalence of sleep problems. Significant differences were noted in problems with parasomnias (a frequent problem for 79% of the children with ASD), sleep onset (43%) and day-time sleepiness (64%).

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Sleep problems in children with Autism Spectrum Disorders (ASD) are commonly reported by their parents (Cortesi, Giannotti, Ivanenko, & Johnson 2010; Didden & Sigafos 2001; Höglund Carlsson et al. 2013; Richdale & Schreck 2009; Wiggs & Stores 2004) and are among the most prevalent comorbid conditions experienced by children with ASD (Xue Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wagner 2007). Comparison studies have reported that children with ASD experience worse sleep than their typically developing (TD) peers (Allik, Larsson, & Smedje 2006; Elrod & Hood 2015; Richdale & Schreck 2009). Estimates for the prevalence of sleep problems in children with ASD vary from 44-83%, in comparison to only 9-50% in TD comparison groups (Elrod & Hood 2015; Richdale & Schreck 2009). Patzold, Richdale and Tonge (1998) noted problems with sleep onset and maintenance as particularly widespread, but also significant levels of sleep-disordered breathing and parasomnias. Sleep problems are not only more prevalent in ASD, but also vary systematically with autistic symptomology in high functioning groups, such that severity of ASD symptoms predicts poor sleep (Hoffman et al. 2005; Schreck 2004).

Methods for measuring sleep in children have developed from parent report via questionnaires or diaries, to the use of direct measures gained from polysomnography and actigraphy. Whilst initial evidence from parent report and diaries helped identify genuine parent concern, parent report has been thought to allow for significant overestimating of sleep problems (Goodlin-Jones, Tang, Liu, & Anders 2008; Hering, Epstein, Elroy, Iancu, & Zelnik 1999). One proposal has been that sleep is actually similar in children with and without ASD (Schreck & Mulick 2000), with differences being a manifestation of the overall strain on parents of children with ASD (Hering et al. 1999; Wiggs et al. 2005). Measuring sleep directly is likely to provide a more accurate representation of the prevalence and severity of sleep problems in children with ASD (Goldman et al. 2009).

In spite of often sharing a single diagnosis (particularly under new DSM-V criteria, American Psychiatric Association 2013), children with ASD are far from being a homogeneous group and many experience comorbid conditions. Most notably, 44-70% of people with ASD also have a comorbid intellectual disability (Fombonne, Quirke, & Hagen 2011; La Malfa, Lassi, Bertelli, Salvini, & Placidi 2004). This is particularly relevant to sleep because of the documented relationship between sleep problems and intellectual disability (Bartlett, Rooney, & Spedding 1985; Berkman 2006; Quine 1992; Richdale, Francis, Gavidia-Payne, & Cotton 2000; Richdale & Baker 2014; Surtees, Oliver, Jones, Evans, & Richards under review; Tietze et al. 2012). With this in mind, one hypothesis is that increased sleep problems identified in the broader ASD population are an artefact of the increased number of children with intellectual disability in this group. Alternatively, there is some evidence from parent report data that suggests that sleep problems are more prevalent in children with ASD (of varying cognitive abilities) than samples of children with developmental delay (Krakowiak, Goodlin-Jones, Hertz-Picciotto, Croen, & Hansen 2008). This conflict makes measuring sleep of children with ASD and no comorbid intellectual disability directly particularly important.

Current evidence from direct measurement of sleep in ASD

A recent systematic review (Elrod & Hood 2015) identified studies that used direct measures to compare sleep in children with and without ASD. Studies reviewed included those using polysomnography (in six cases), actigraphy (in four cases) and one study in which both were employed. Polysomnography measures brain activity through electroencephalography, oxygen saturation in the blood, respiratory rate, heart-rate and movement and is considered the gold standard within sleep research (Michaelson, Allan, Chaney, & Mair 2006), but is

limited by measuring across a short time period. In all but one study in children with ASD (Goldman et al. 2009), only a single night was used in the final analysis. Studies employing actigraphy measure only movement, typically with a small watch-like device on the wrist. Actigraphy is considered less accurate than polysomnography, particularly because of its poor sensitivity in identifying restful waking periods (Sadeh & Acebo 2002). Conversely, actigraphy is easier to employ for longer periods (Allik et al. 2006; Goodlin-Jones et al. 2008; Souders et al. 2009; tested for the recommended five nights or more) and for testing in the child's natural environment. Restricted preferences are a diagnostic feature of ASD, predictive of specific difficulties in adapting to new environments. With this in mind, employing actigraphy, as recommended for 5-7 nights in the child's home, may be a more sensitive and accurate measure of typical sleep patterns for children with ASD. However, polysomnography would, of course, remain more valid for identifying time spent in different stages of sleep, apnoea, sleep-disordered breathing and other parasomnias.

Elrod and Hood's (2015) meta-analysis of comparisons between sleep in children with ASD and TD controls showed that across the 11 studies, there were significant differences between children with ASD and TD on Total Sleep Time (on average 32.8 minutes per day shorter in ASD), Sleep Latency (10.9 minutes longer per day in ASD) and Sleep Efficiency (1.9% per day higher per day in ASD). Elrod and Hood (2015) also tested whether the effect was moderated by the method used or the exclusion of children with intellectual disabilities, children on medications or children with seizure disorders. The only significant moderator identified was the effect of excluding children with intellectual disabilities on Total Sleep Time. When the analysis only included the three studies which did not include children with intellectual disabilities (two using polysomnography and one actigraphy), there were no significant differences in Total Sleep Time. This finding is perhaps surprising, given the well-documented relationship between ASD and insomnia

(Richdale & Shreck 2009). There were differences identified in Sleep Latency and Sleep Efficiency, but these could be explained by parents of children with ASD setting earlier bedtimes for their children¹ (Allik et al. 2006). Alternatively, parents of children with ASD may *have* to set earlier bedtimes to achieve equivalence of Total Sleep Time *because* of long sleep latencies and poor sleep efficiency. With this in mind, an actigraphy study measuring the sleep of children with parent-identified sleep problems is timely.

Allik et al. (2006) completed the only published study employing actigraphy for five to seven nights for a group of children with ASD and no comorbid intellectual disability (Goldman et al. 2009, only employed actigraphy for two nights). Nineteen children with Asperger's Syndrome and 13 with High-Functioning Autism (age range eight to 13) were recruited pseudo-randomly from specialist services in Sweden and compared to 32 TD controls. Interestingly, in spite of not recruiting children with intellectual disabilities, the majority of the children with ASD still attended "classes or schools for children with various special needs" (Allik et al. 2006, p588). Within the sample of children with ASD, 59.2% were reported to have sleep problems, in comparison to only 9% in the control group. Consistent with Elrod and Hood's (2015) meta-analysis, they identified no significant difference in Total Sleep Time: averaging 511 minutes (on school days) to 514 minutes (on weekends) for children with ASD, in comparison to 523 (weekend) to 522 (weekday) minutes for controls. However, children with and without ASD differed on both Sleep Efficiency: 87% for ASD, 90% for controls, and Sleep Latency: 22-32 (weekday-weekend) minutes for ASD, 11-16 (weekday-weekend) minutes for TD. Whilst Allik et al.'s study (2006) provides evidence for similarities and differences between ASD with no concurrent intellectual disability and TD controls, there remain limitations. Firstly, there was no evidence provided

¹ If bedtime is set earlier, children may take longer to get to sleep and spend longer in bed overall, thus decreasing sleep efficiency.

of the children's scores on cognitive testing (though these were assessed in patient notes), on a standardized assessment for ASD or on a measure of adaptive functioning. This limits understanding of how representative of the broader ASD population the group were and does not allow for quantifying how autistic symptomology, cognitive abilities or adaptive functioning may have related to sleep. Secondly, the authors do not report what proportion of children may have experienced a specified sleep problem. Finally, it is difficult to draw firm conclusions from a single study of this kind.

Explaining poor sleep in ASD

Whilst researchers broadly agree on the increased prevalence of sleep problems in children with ASD, the mechanism underlying these problems is less clear (Cortesi et al. 2010). Richdale and Schreck (2009) proposed a model that emphasises biological, psychological *and* social factors that predispose children with ASD to experiencing sleep problems and further perpetuate them. *Biological* mechanisms proposed include circadian rhythm dysfunction, irregular expression of clock genes and abnormal melatonin release or synthesis (Bourgeron 2007; Richdale & Schreck 2009). Importantly, it has been noted that night-time melatonin levels were inversely related to severity of autistic symptoms (Tordjman, Anderson, Pichard, Charbuy, & Touitou 2005).

Proposed *psychological* mechanisms have mainly focussed on the psychological and behavioural features of ASD: deficits in communication, social interaction and restricted interests/ repetitive behaviours. This hypothesis is supported by the evidence that poor sleep correlates not only with overall ASD symptomatology (Hoffman et al. 2005; Malow et al. 2006; Schreck et al. 2004), but also communication (Quine 1992), social skills (Hoffman et al. 2005) and stereotypic behaviours (Schreck et al. 2004) independently. There is little

evidence, however, for the direction of causality of this effect and poor sleep could precipitate an increase in symptom severity. One possibility is that sleep problems exacerbate difficulties in ASD symptoms (Schreck et al. 2004), perhaps mediated through established links between poor sleep and executive function (Jones & Harrison 2001). Another is that poor sleep may be precipitated by increased challenging behaviour associated with more severe ASD symptoms (Wiggs & Stores 1996). ASD symptomology is also confounded with intellectual disability, which predicts poor sleep, so may mean that the association between ASD symptomology and sleep is in no way causal. Further psychological mechanisms proposed have included potential comorbidities with mood disorders, such as anxiety and/ or depression (Cortesi et al. 2010) and Attention Deficit/ Hyperactivity Disorder (ADHD; Liu, Hubbard, Fabes, & Adam 2006).

The contribution of social mechanisms to sleep problems has received less attention in the literature (Richdale & Schreck 2009). Richdale (1999) noted that children with ASD and sleep problems often have unusual sleep routines, particularly around settling at bed-times. There is also good evidence that parents of children with ASD experience high levels of stress (higher even than parents of children with other disabilities; Dunn, Burbine, Bowers, & Tantleff-Dunn 2001) and parent stress levels are correlated with poor sleep (Richdale et al. 2000); though again direction of causality is unclear.

Rationale

A body of research has examined the prevalence, severity, nature and cause of sleep problems in children with ASD. However, very few of these studies have measured sleep directly, through actigraphy or polysomnography. Most studies that have done so have included children with a range of cognitive and functional abilities. On the one hand this is important

in understanding sleep across the broader population of children with ASD. Conversely, it is difficult to identify the independent and complimentary roles for intellectual disability and ASD in predicting the poor sleep. The current study used actigraphy to compare sleep in children with ASD to a TD comparison group. Only children with a parent-reported sleep problem were recruited to the ASD group to understand the nature of these problems (rather than estimate the prevalence of sleep problems in people with ASD more broadly). None of the participants tested had a comorbid intellectual disability, defined to require a performance IQ < 70. Parent diaries were also undertaken to compare parent reports of sleep in the two groups and evaluate their accuracy using actigraphy. Sleep questionnaires investigated parent reports of specific problems with sleep. All children with ASD were tested using direct measures of IQ, adaptive behaviour and a range of questionnaires, to describe robustly the participant characteristics and identify the relationship between poor cognitive functioning, adaptive behaviour, daytime functioning and sleep.

Aims

This study compared children with ASD who had a parent reported sleep problem and typically developing children in three key areas:

- I. Similarities and differences in sleep, measured directly by actigraphy.
- II. Similarities and differences in parent reports of sleep.
- III. Frequency of different parent-reported sleep problems.

Data from children with ASD were further examined to investigate:

- IV. Similarities and differences between actigraphy measures and parent reports.
- V. Individual differences in sleep quantity and quality using correlations.

Method

Participants.

Sixteen children (see table 1) with Autism Spectrum Disorders (mean age = 9.8; 63% male; average performance IQ = 100.7; average adaptive behaviour standard score = 77.4) were matched on age ($t = .83$; $p = .22$) and gender to 16 typically developing children. Post-hoc comparisons showed no differences between the two groups on maternal education ($\chi^2 = 6.95$, $p = .07$) or family income grouping ($\chi^2 = 6.79$, $p = .45$). All families of children with ASD on a variety of databases² were contacted and invited to take part if their children fulfilled three criteria: an existing diagnosis of ASD, a current sleep problem (decided by parents) and being aged five-15. Three further children were recruited to the original sample, but were excluded from this study. Two of these children were excluded because sleep diaries were incomplete. The final child was excluded after scoring below the normal range on cognitive testing. The final sample included four families in which more than one child with ASD took part (total $N = 10$). Parents of two children in the final sample did not return questionnaires within the identified time period and so are not included in this part of the analysis.

Typically developing children were recruited through contacts of researchers and students at the university. A larger sample of 44 children were recruited, with the final sample selected to match for age and gender. Typically developing children were excluded if parents reported that they had a statement of special educational needs which indicated that had a condition that may have impacted upon their sleep quality (e.g. attentional difficulties

² Databases included: A local area database of children with ASD in the West Midlands (UK), a research centre database (including children with a variety of genetic syndromes and developmental disorders), a database of children with ASD from a second research group and a small database of parents who attended a charity-led workshop on sleep problems in developmental disorders.

and hemiplegia). All parents of typically developing children selected scored below the ASD cut-off on the Social Communication Questionnaire (Rutter, Bailey & Lord, 2003; indicative that none of them were likely to have ASD).

A greater number of parents of children with ASD reported that their children had experienced health problems in the month prior to testing (ten vs. five). In line with being recruited based on having a sleep problem, more children in the ASD group were currently taking sleep medication (five vs. zero)³.

(Please insert table 1 near here)

Procedure

On recruitment, children with ASD and their parents attended a research centre for direct assessments of cognitive abilities and autistic symptomology. An assessment of adaptive behaviour was completed with parents by interview over the telephone. All assessments were completed in the 15 weeks prior to the week in which sleep was measured directly using actigraphy. Direct assessments of IQ, autistic symptomology and adaptive behaviour were not completed for typically developing children. All children in the typically developing comparison group attended mainstream primary or secondary schools.

³ Removing children on sleep medication from the primary analyses did not affect the significance of any results.

Assessment of ASD, intellectual and adaptive functioning

Diagnosis of Autism/ Autism Spectrum Disorder was corroborated by completion of the Autism Diagnostic Observation Schedule (ADOS; Lord et al. 2000). To provide consistency of testing across a sample that varied in age and were initially of unknown cognitive ability, all children completed the Mullen Scales of Early Learning (Mullen 1995). All children who achieved ceiling scores on the Mullen were tested using the performance subscales of the Wechsler Abbreviated Scale of Intelligence (Wechsler 1999). One five-year old boy was below the minimum age suggested for the WASI, but scored above average for a six-year old on the test, suggesting he had no cognitive impairment (his performance IQ was not included in examination of individual differences in sleep). The Vineland Adaptive Behavior Scales (Sparrow et al. 1984) was used to measure adaptive behaviour and functioning.

Actigraphy.

Each child wore an Actiwatch 2 (Phillips Respironics) on the wrist for a continuous period of seven to eight days, in line with Acebo et al.'s (1999) guidance on obtaining reliable measures of sleep through actigraphy. Children and their parents were directed that, if possible, the watch should be worn at all times. Data were measured in 30 second epochs. Sleep intervals were calculated automatically using Actiware software (version 6.0.7) using the default medium sensitivity threshold for night waking. The start and end of the sleep intervals were identified by the start and end of 10 minutes of continuous epochs scored as sleep respectively. Data cleaning was undertaken to remove artefacts that can make actigraphy data unreliable (Acebo et al., 1999). Sleep intervals were altered if the watch was removed or the interval missed a significant period of sleep within the child's reported time in bed. Variables extracted from the actigraphy measure included Bed-time (BT; the time at which children entered a restful state), Get-up time (GT; the end time of the final period of

sleep in the morning), Onset Latency (OL; the time between BT and the first period encoded as sleep), Wake After Sleep Onset (WASO; The amount of time after first period of sleep spent awake), Time in Bed (TiB; the time between BT and GT), Total Sleep Time (TST; the recorded time spent asleep each night) and Sleep Efficiency (SE; the percentage of TiB recorded as sleep).

Diary measures.

Parents completed a diary for the period over which sleep was measured. This diary included questions about their child's sleep: time they went to bed, time lights were turned out, time parents felt their child awoke, time they got out of bed, time they took to get to sleep, day-time naps, night-time awakenings, difficult behaviours around bed-times and also details of their own interventions to promote sleep. Variables were calculated from diaries to match those from actigraphy. In most cases, this was transposed directly from parent report. In addition, three composite variables were calculated: Time in bed = Time out of Bed – Bed Time; Total Sleep Time = Wake-up Time – Lights Out Time – Total Waking Time – Time to get to Sleep; Sleep Efficiency = $100 \times (\text{Total Sleep Time} / \text{Time in Bed})$.

Questionnaires.

Parents completed a pack of questionnaires within a week of direct assessment of sleep. For background, all parents completed questionnaires on demographic and health information. All parents completed the Social Communication Questionnaire (Rutter et al. 2003) to measure ASD symptomology in children with ASD and exclude children with potential ASD symptomology in the group of typically developing children. All parents completed a range

of sleep questionnaires to examine group differences and correlate with direct measurement of sleep.

Modified Simonds and Parraga Sleep Questionnaire (Simonds & Parraga 1982; Wiggs & Stores 2004)

The Simmonds and Parraga Sleep Questionnaire is a broad ranging parent-report measure of sleep in children (Simmonds & Parraga 1982). The modified version was adapted for children with developmental disabilities (Wiggs & Stores 2004). The measure can be used to calculate an overall measure of sleep problems (Johnson, Turner, Foldes, Malow, & Wiggs 2012), which correlates well with the Childhood Sleep Habits Questionnaire (Owens et al. 2000). It can be broken down into seven subscales: Bed-time Resistance, Sleep Onset Delay, Night Wakings, Sleep Anxiety, Parasomnias, Sleep Disordered Breathing and Daytime Sleepiness.

Obstructive Sleep Apnoea (Mindell & Owens, 2003)

The Obstructive Sleep Apnoea Questionnaire (Mindell & Owens 2003) is used as a screening measure, to consider whether children require further assessment. It contains both broad level questions about sleep and sleepiness, such as “Is it hard to wake your child up in the morning?” and more specific apnoea-related questions, such as “When sleeping does your child have trouble breathing, or struggle to breathe?”. All items require yes/no responses and an average score (0-1) calculated, with a cut-off advised at $>.33$.

Family Inventory of Sleep Habits (Malow et al. 2008)

The Family Inventory of Sleep Habits (FISH, Malow et al. 2008) is a broad measure of sleep hygiene, focussing on an individual child's routine. The questionnaire was developed to assess sleep hygiene in children with ASD.

Modified Paediatric Epworth Sleepiness Scale (Williams, Scheimann, Sutton, Hayslett, & Glaze 2008).

The Epworth Sleepiness Scale (Epworth 1991) measures daytime sleepiness through asking people how likely they are to “doze” in a range of situations. The modified paediatric version (Williams et al. 2008) differs in asking for a parent response and in removing situations that are less likely to be experienced by children (such as having drunk alcohol).

Data Analysis

Group Comparisons and correlations

Outcome variables from actigraphy, sleep diaries and questionnaires were compared between the children with ASD and the TD comparison group using independent samples t-tests. Relationships between actigraphy measures and parent report were analysed using Pearson's correlations. The relationship between individual differences in sleep time and sleep efficiency, and other variables were examined using Spearman's correlations due to some questionnaire data differing significantly from a normal distribution. For all statistical tests, $p < .01$ was used for significance to accommodate multiple comparisons with an acceptable risk of type-1 and type-2 error. Results on which $.01 \leq p < .05$ were considered trends to lower the risk of not reporting potentially clinically significant results.

Results

Data were analysed to address each of the aims of the study. Differences in sleep between the groups of children with and without ASD were investigated by analysing differences in actigraphy data. Differences in parent reports of sleep between the groups of children with and without ASD were tested using data from sleep diaries. Frequency of different parent-reported sleep problems in the two groups were identified using the sleep questionnaires and their subscales. Similarities and differences between actigraphy measures and parent reports were then analysed. Individual differences in sleep quantity and quality were identified by correlating outcome variables from actigraphy with demographics and questionnaire totals.

Group differences in sleep, measured by actigraphy.

Independent samples t-tests were undertaken comparing the two groups on each of the dependent variables taken from actigraphy. No group differences on any of the measures were identified, see table 3.

(Please insert table 2 near here)

Group differences in parent reports of sleep

To compare parent reports of sleep time in their children, responses from the sleep diaries of the two groups were compared. Again, independent samples t-tests found no significant differences between the groups (see table 3). There were trends for the children with ASD being reported to wake up and get out of bed earlier than the TD children.

(Please insert table 3 near here)

Frequency of parent-reported sleep problems in children with and without ASD.

To investigate the frequency and nature of parent-reported sleep problems, independent samples t-tests compared the two groups' scores on each of the questionnaires. Children in the ASD group scored significantly higher than children in the TD group on both measures related to sleep itself (the MSPSQ and the OSAQ; see table 4). There was no evidence of group differences in sleep hygiene or day-time sleepiness (as measured by the FISH and the MPRESS).

(Please insert table 4 near here)

Specifying Sleep Problems

Whilst diagnosing a sleep problem requires more in-depth clinical information than available in this study, the MSPSQ does afford the opportunity for identifying potential areas of concern. Johnson et al. (2012) suggest 56 as a cut-off on the MSPSQ total score as indicative of a sleep problem. Using this cut-off, 12 of 14 children (86%) with ASD were considered to have a sleep problem and only five of 16 typically developing children (31%). This represented a significant difference between groups ($X^2(1) = 9.02$ $p = .004$).

To investigate the likely sleep problems responsible for group differences on the MSPSQ, the subscales of the questionnaire (Johnson et al. 2012) were investigated. There were significant differences between the groups, such that the children in the ASD group scored higher than those in the TD group on Sleep Onset Delay ($t(1) = 2.89$, $p = .007$), Parasomnias ($t(1) = 3.22$, $p = .003$) and Day-Time Sleepiness ($t(1) = 3.44$, $p = .002$) and there was a trend for a difference on Night Waking ($t(1) = 2.68$, $p = .012$), see fig 1. Wiggs & Stores (2004) suggest that any score above four on items of the MSPSQ is suggestive of a frequent problem that may be cause for concern. Using this criterion, 79% of the children with ASD evidenced potential problems with parasomnias, 43% with sleep onset and 64%

with day-time sleepiness (compared to 13%, 19% and 19% respectively in the TD group).

There were no significant differences on other subscales ($ts(1) \leq 1.87, p \geq .07$).

(Please insert fig. 1 near here)

Similarities and differences between actigraphy measures and parent reports.

Pearson's correlations were employed to investigate the relationship between equivalent variables measured by actigraphy and sleep diaries in children in the ASD group. There were significant positive correlations between diary reported bed-times, get-up times, times in bed and total sleep times and those found through actigraphy. A trend was identified for equivalent relationships in sleep onset. Paired samples t-tests were used to identify differences between actigraphy and parent report. In most cases, there were significant differences between actigraphy recordings and parent report: Parents reported their children to go to bed earlier, spend longer in bed and get more sleep in total than recorded by actigraphy. Parents also recorded shorter waking times than identified by actigraphy.

In sum, parent reports of sleep time (but not night-time wakings) reflected an accurate representation of individual differences between children. Differences between parent reports and direct measurement reflected parents overestimating their children's sleep time.

(Please insert table 5 near here)

Individual differences in Sleep Quantity and Quality

Demographic information and totals from the questionnaires were correlated against Total Sleep Time and Sleep Efficiency measures from actigraphy (table 6). Spearman's Rank correlations were used as some questionnaire data differed significantly from the normal distribution. There were no significant correlations, but there were trends for a negative correlation between Sleep Time and Age and between adaptive behaviour and sleep efficiency.

(Please insert table 6 near here)

Discussion

The sleep of children with ASD was compared to that of typically developing children using a comprehensive range of measures. Recruitment to the ASD group required parents considering their child to have a sleep problem, whilst typically developing children were not recruited under this condition. The most objective measurements of sleep, through actigraphy, found no significant differences between the two groups. Parent reports correlated well with these objective measurements, other than for wakings, and also showed no significant group differences. On questionnaire measures, however, there was evidence of group differences in sleep. Notably, children in the ASD group returned significantly higher scores on both the general sleep questionnaire (the MSPSQ; Wiggs & Stores 2004) and the Screening Questionnaire for Sleep Apnoea (Mindell & Owens 2003).

Measuring Sleep Directly through Actigraphy

Consistent and reliable reports have concluded that sleep problems are more common in children with ASD (44-83% of children with ASD experience sleep problems), than they are in the general population (9-50% of children more generally; Elrod & Hood 2015; Richdale & Schreck 2009). It is therefore surprising that measurements of children's sleep taken through actigraphy were not significantly different for children with ASD from children who were matched in age and gender, but did not have ASD. Such a conclusion is even more stark given that the children with ASD were recruited on the basis of a parent-reported sleep problem. Over a week of recordings, no significant differences between groups were observed in duration of sleep (as measured through Total Sleep Time) and quality of sleep (as measured through Wake After Sleep Onset and Sleep Efficiency). These findings are not entirely inconsistent with previous findings in children with ASD *without* comorbid intellectual disabilities. A recent meta-analysis (Elrod & Hood 2015) showed that the three studies that measured sleep directly, through polysomnography or actigraphy, found no overall difference in duration of sleep between children with and without ASD (and no comorbid intellectual disability). The findings here are consistent with that result. The meta-analysis (Elrod & Hood 2015) did, however, find significant differences in sleep efficiency and sleep latency between groups. Here these were not observed.

The only other study using actigraphy appropriately in children with ASD without comorbid intellectual disabilities (Allik et al. 2006) similarly found no group differences in overall sleep time, but did evidence differences in latency and efficiency. Interestingly, both groups in the study by Allik and colleagues (Allik et al. 2006), returned higher estimates of sleep efficiency (in spite of being of similar ages) than in this study. It is therefore possible that the control group here were unusually poor sleepers. Typically developing children were recruited through opportunity sampling by university staff and students, with social media

being commonly used. It is possible that parents of children with irregular sleep patterns may have been more attracted by the opportunity to receive detailed feedback on their child's sleep (though note questionnaires suggested parents of children with ASD reported significantly more sleep problems than did those of TD children).

Parentally Reported Diaries

Parental reports of the sleep of their children with ASD correlated well with measures gained through actigraphy. Interestingly, comparisons between the two measures showed that parents actually overestimated their children's sleep duration and efficiency. Evidence suggested that this was the result of underestimating the time their children were awake after going to sleep for the first time. These results suggest that parents of children with ASD were regularly unaware of the times at which their children were awake. Whilst direct measures of sleep are often preferred on the basis that parents may overestimate their children's sleep difficulties (Goodlin-Jones, Tang, Liu, & Anders 2008; Hering, Epstein, Elroy, Iancu, & Zelnik 1999), in this study, there is evidence for the opposite. This stands as further evidence for the preference of direct measures in both research and clinical practice. There were no significant differences between groups on diary-reported measures, although there were trends towards children with ASD going to bed earlier and rising earlier than their typically developing peers. As for actigraphy, it was surprising that data from children with ASD and a reported sleep problem did not significantly differ from that from their peers. This suggests that parents did not believe their child to have a sleep problem based on underestimating their sleep duration. One explanation is that they may have had an unrealistic expectation of the sleep their child should be getting. Another is that parental experiences of sleep problems

included broader difficulties with sleep than duration of sleeping and waking. For instance, parental experiences may, reflect the difficulties their child presents with on waking.

Questionnaires

Unlike measures from actigraphy and parent-report diaries, analysis of questionnaires did show differences between the two groups. This was consistent with children in the ASD group being recruited on the basis of a reported sleep problem. Most notably, the children with ASD scored significantly higher on the MSPSQ (Wiggs & Stores 2004) and the OSQ (Mindell & Owens 2003). Both of these questionnaires assess sleep and night-time behaviour much more broadly than the measures collected in the diaries and measured through actigraphy. The MSPSQ (Wiggs & Stores 2004), for instance, includes questions on bed-time resistance, day-time sleepiness and parasomnias, as well as questions on sleep latency and night-time waking. The OSQ (Mindell & Owens 2003) includes items thought to be indicative of sleep apnoea, such as whether a child stops breathing at night, but also items associated with day-time sleepiness or inattention.

Subscales on the MSPSQ showed significant group differences on Sleep Onset, Parasomnias and Daytime Sleepiness. Diaries and actigraphy showed no difference in sleep latency, so differences in the sleep onset subscale were surprising, perhaps suggesting they reflected differences in parental perception. Similarly, differences in day-time sleepiness may be surprising as they were not identified on the broader scale of sleepiness (The Pediatric Epworth Sleepiness Scale). One reason may be that the Epworth scale focuses on a single criterion of the “likelihood of dozing”, which may not reflect the broader experience of sleepiness. Differences on the parasomnias subscale were informative and report aspects of sleep that may not have been identifiable through actigraphy. Notably nearly 80% of the children with ASD experienced at least one form of parasomnia once a week or more (in

comparison to less than 15% of the control group). Though dyssomnias (difficulties in initiating or maintaining sleep) have received more attention in the literature, there is previous evidence suggestive of parasomnias being more prevalent in children with ASDs (Gail Williams, Sears, & Allard 2004; Ming, Sun, Nachajon, Brimacombe, & Walters 2009). In the only polysomnography study of parasomnias in ASD, Ming et al. (2009) reported a particularly high prevalence of disorders of partial arousal in children with ASD. Partial arousals may indicate poorer sleep and pre-dispose children to sleep terrors and confused awakenings. Though cautioned by the possibility that children with ASD may have been more influenced by testing in a sleep laboratory, Ming et al. (2009) suggested that one reason for this could be greater fragmentation in sleep more generally. Further research in this area may be able to define more clearly the precise nature and prevalence of parasomnias in children with ASD, their likely biopsychosocial precipitants and perpetuators and their impact on day-time functioning.

One caveat is that questionnaire measures of sleep in children can be criticised for measuring parent expectations as well as the child's actual sleep patterns. Day-time behaviour may impact parental stress and thus make sleep problems seem more severe. In this study, one reason to believe this may not have been the case was the reliability of parent-reports on the sleep diaries. Parents overestimated their children's sleep duration, but reported further sleep problems alongside this.

Limitations and future directions

Although a direct measure of sleep, actigraphy has significant limitations in comparison to polysomnography (Michaelson, et al. 2006). Actigraphy can often misrepresent restful waking as sleep (Sadeh & Acebo 2002). It can also miss finer-grained distinctions in sleep

cycles, such as time spent in REM sleep and evidence for sleep apnoea, both of which can be accurately recorded using polysomnography. The finding from this sample that estimates of parasomnias were high suggests that polysomnography may have more accurately represented the concerns parents had about their children's sleep. Only two studies have compared sleep in children with ASD and no comorbid intellectual disability using polysomnography and each of these only recorded from a single night. Larger samples and longer testing periods of polysomnography may be needed to understand sleep problems in this group.

The study was also limited by sample size and nature. Only 16 children with ASD were recruited and these were drawn from a relatively broad age range and a broad range of cognitive and adaptive abilities (though all were in the normal range on a standardized measure of performance IQ). That all children were recruited on the basis of their parents reporting them to have a sleep problem, limits the study in describing sleep problems in children with ASD, rather than a broad population of children with ASD.

Summary

Sleep problems in children with ASD have been well-documented and are cited as among the most common comorbid conditions for this group (Xue Ming et al. 2007). There have, however, been very few studies measuring sleep using direct measures that have compared sleep in children with ASD and no comorbid intellectual disability to children without ASD. Here, actigraphy was used to add to that literature. In support of a recent meta-analysis (Elrod & Hood, 2015), there was no evidence that the children with ASD, with no intellectual disability, slept for shorter periods. Interestingly, even though children were recruited on the basis of their parents considering them to have a sleep problem, there was no evidence of greater durations of waking or longer sleep latencies either. Diary measures suggested that

parents were not underestimating the actual sleep their children got or overestimating their waking periods. Questionnaire data suggested that sleep problems in children with ASDs may reflect a broader range of sleep difficulties.

Compliance with Ethical Standards:

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Ethical approval: All procedures performed in this study were in accordance with local ethical guidelines and with the 1964 Helsinki declaration and its later amendments.

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Table 1. Participant characteristics of children with ASD recruited for the study

Participant	Age (Years)	Gender	Performance IQ ¹	Adaptive Behaviour ²
1	12	Female	112	68
2	7	Female	95	89
3	8	Male	131	75
4	13	Male	89	76
5	14	Male	94	79
6	11	Female	111	78
7	8	Male	110	80
8	12	Male	110	74
9	10	Male	112	62
10	9	Male	70	68
11	10	Male	76	66
12	7	Female	77	126
13	11	Female	112	68
14	10	Male	127	97
15	5	Male	>103	65
16	10	Female	84	67

¹Score on the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999)

²Standard Score on the Vineland Adaptive Behavior Scales (Sparrow et al., 1984)

Table 2. Group mean scores and differences for measurements from actigraphy. * $p < .05$

	ASD Mean (SD)	TD Mean (SD)	<i>t</i>	<i>p</i>
Bed Time (hh:mm)	20:46 (1:01)	21:26 (1:05)	1.75	.09
Get-up Time (hh:mm)	06:46 (0:59)	07:15 (0:35)	1.70	.10
Time in Bed (hh:mm)	09:59 (0:46)	09:49 (1:04)	.51	.62
Total Sleep Time (hh:mm)	08:05 (0:39)	08:12 (0:45)	.65	.62
Onset Latency (hh:mm)	00:38 (0:32)	00:29 (0:26)	.19	.37
Sleep Efficiency (%)	82.08 (4.17)	83.88 (5.68)	1.02	.32
Wake After Sleep Onset (hh:mm)	0:52 (0:17)	00:48 (0:16)	.68	.50

Table 3. Group mean scores and differences for measurements from sleep diaries. * $p < .05$

	ASD Mean (SD)	TD Mean (SD)	<i>t</i>	<i>p</i>
Bed Time (hh:mm)	20:25 (0:58)	20:55 (0:58)	1.44	.16
Lights Out (hh:mm)	20:51 (1:05)	21:20 (1:09)	1.26	.22
Wake up time (hh:mm)	06:39 (0:47)	07:12 (0:46)	2.05	.05*
Time out of Bed (hh:mm)	06:52 (0:49)	07:31 (0:45)	2.30	.03*
Time to get to Sleep (hh:mm)	0:39 (0:29)	0:32 (0:19)	.81	.43
Wake After Sleep Onset (hh:mm)	0:11 (0:17)	0:03 (0:03)	1.77	.09
Time in Bed (hh:mm)	10:28 (0:51)	10:24 (0:49)	.16	.87
Total Sleep Time (hh:mm)	9:03 (1:07)	9:16 (1:22)	.45	.65
Sleep Efficiency (%)	86.5% (8%)	88.7% (10%)	.70	.49

Table 4. Group mean scores and differences for questionnaires. ** $p < .01$

Variable	ASD Mean (SD)	TD Mean (SD)	<i>t</i>	<i>p</i>
Modified Simonds and Parraga Sleep Questionnaire	73.05 (15.16)	53.29 (13.59)	3.77	.001**
Obstructive Sleep Apnoea Questionnaire	.41 (.14)	.13 (.09)	6.73	< .001**
Family Inventory of Sleep Habits	50.57 (11.06)	46.60 (5.02)	1.26	.22
Modified Paediatric Epworth Sleepiness Scale	2.21 (3.02)	2.50 (1.97)	.31	.76

Table 5. Correlations and comparisons between actigraphy and Sleep diary measures for children with ASD (See appendix 1.2.3 for equivalent statistics in the TD group). Means are contained in tables 1.2.1 and 1.2.2. ** $p < .01$, * $p < .05$.

Actigraphy Variable	Sleep Diary Variable	<i>R</i>	<i>p</i>	<i>t</i>	<i>p</i>
Bed Time	Bed Time	.94	< .001**	3.98	.001**
Get up time	Wake Time	.92	< .001**	1.05	.31
Time in Bed	Time in Bed	.81	< .001**	3.74	.002**
Total Sleep Time	Total Sleep Time	.68	.003**	4.74	< .001**
Onset Latency	Time to get to sleep	.50	.048*	.16	.87
Sleep Efficiency	Sleep Efficiency	.15	.57	2.05	.06
Wake After Sleep Onset	Wake After Sleep Onset	-.03	.90	6.80	< .001**

Table 6. Correlation between dependent variable questionnaires and sleep outcome measures (from actigraphy) in the ASD group (See supplementary materials for the equivalent statistics from typically developing children). * $p < .05$.

Measure	Correlation with Total Sleep Time, ρ (p)	Correlation with Sleep Efficiency, ρ (p)
Age	-.50 (.05*)	-.31 (.25)
Performance IQ	.07 (.80)	-.23 (.42)
Adaptive Behaviour (VABS)	.15 (.59)	-.54 (.03*)
Sleep Problems (MSPSQ)	.28 (.33)	-.03 (.91)
Sleepiness (PESS)	-.05 (.88)	-.11 (.72)
Social Communication (SCQ)	.09 (.78)	.06 (.84)
Family Sleep Habits (FISH)	.22 (.46)	.23 (.42)

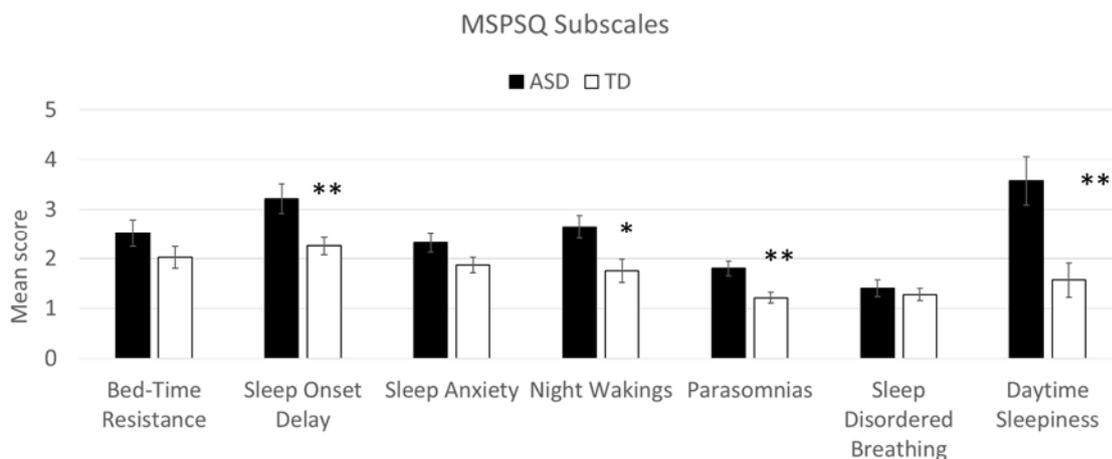


Figure 1. Comparisons between groups on subscales of the Modified Simmonds and Parraga Sleep Questionnaire. Error bars indicated standard error of the mean. ** $p < .01$, * $p < .05$.