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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

Background: It has been reported widely that children with Autism Spectrum Disorders (ASD) are more likely to experience sleep problems than children without ASD. Sleep problems are among the most prevalent comorbid difficulties with ASD. The current study aimed to use multiple methods to describe these difficulties.
Method: Sleep of sixteen children with ASD and a parentally-reported sleep problem was compared to the sleep of a matched group of children without ASD. Seven nights of actigraphy data were collected for both groups, alongside sleep diaries and questionnaires.
Results: 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%).
Conclusions: Multi-method assessment is vital in understanding sleep problems in children with ASD. Broad estimates of quantity of sleep do not necessarily describe the difficulties experienced. Using questionnaires in addition to objective measurement may be a means to understand sleep problems in children with ASD and to an improved understanding of their impact.

1. Introduction

Sleep problems in children with Autism Spectrum Disorders (ASD) are commonly reported by children themselves (Baker, Richdale, Short, & Gradisar, 2013; Paavonen et al., 2008; Richdale & Baglin, 2015) and by their parents (Cortesi, Giannotti, Ivanenko, & Johnson, 2010; Didden & Sigafoos, 2001; Höglund Carlsson et al., 2013; Richdale & Schreck, 2009; Wiggs & Stores, 2004). They are among the most prevalent comorbid conditions experienced by children with ASD (Ming, Brimacombe, Chaaban, Zimmerman-Bier, & Wager, 2007). Understanding the topography of sleep in these children with ASD and a sleep problem is therefore of crucial importance.

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Estimates for the prevalence of sleep problems in children with ASD vary from 44 to 83%, in comparison to only 9–50% in TD comparison groups (Elrod & Hood, 2015; Richdale & Schreck, 2009). Patzold, Richdale, & Tonge, 1998) noted problems with sleep onset and maintenance as particularly widespread. Sleep problems are not only more prevalent in ASD, but also vary systematically with autistic symptomology: the severity of ASD symptoms predicts poor sleep (Hoffman et al., 2005; Schreck, 2004). 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).

In spite of often sharing a single diagnosis (particularly under DSM-5 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 & Baker, 2014; Richdale, Francis, Gavidia-Payne, & Cotton, 2000; Surtees, Oliver, Jones, Evans, & Richards, 2018; Tietze et al., 2012). Similarly, physical health conditions, such as epilepsy (Tuchman & Rapin, 2002) and mental health conditions such as anxiety and depression (Kim, Szatmari, Bryson, Streiner, & Wilson, 2000) are more prevalent in autistic children and independently predict poor sleep (Batista & Nunes, 2007; Johnson, Chilcoat, & Breslau, 2000).

1.1. Methods for measuring sleep in children

Methods for measuring sleep in children have developed from parent-reported questionnaires or diaries, to combining this with measures gained from objective measures, such as polysomnography and actigraphy. Evidence from parent report and diaries identifies broad features of sleep topography and documents parent concern. Measuring sleep objectively is likely to provide a more accurate representation of the prevalence and severity of sleep problems in children with ASD (Goldman et al., 2009). Polysomnography measures brain activity through electroencephalography, oxygen saturation in the blood, respiratory rate, heart-rate, and movement. Polysomnography is considered the gold standard within sleep research (Michaelson, Allan, Chaney, & Mair, 2006), but is limited as standard protocols involve measuring across a short time period. Typically, this involves a single night of testing preceded by a night of adaptation to the environment/ equipment. Studies employing actigraphy measure only movement, typically with a small watch-like device on the wrist (Sadeh, 2011). 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. Restricted preferences are a diagnostic feature of ASD, predictive of specific difficulties in adapting to new environments. Though polysomnography protocols include a night of adaptation, it is not clear that a single night is sufficient for children with ASD to adapt to a new environment. 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.

The current study uses both objective and subjective measures to compare sleep in children with and without ASD. Specifically, actigraphy is combined with parent-report diaries and questionnaires. Recent systematic reviews and meta-analyses provide a basis for understanding our study.

1.2. Current evidence from subjective measures comparing sleep in children with and without ASD

A recent systematic review (Díaz-Román, Zhang, Delorme, Beggiato, & Cortese, 2018) reviewed papers that compared sleep in young people with and without ASD. Of 47 papers identified, 37 included subjective measures of sleep. Though they included studies with samples of young people $\leq$ 20 years old, all samples had a mean age $<$ 18. A meta-analysis of these results demonstrated poorer sleep in the children with ASD on almost all measures. Children with ASD were reported to have more problems with bedtime resistance, sleep onset delay, night awakenings, parasomnias, sleep-disordered breathing, daytime sleepiness, "restorative value of sleep", sleep onset latency (as estimated in time), and general sleep problems. No difference was identified in sleep quality, sleep efficiency or sleep duration.

1.3. Current evidence from objective measures comparing sleep in children with ASD to their peers

Díaz-Román et al. (2018) also conducted a meta-analysis of data from objective measures of sleep, as did Elrod and Hood (2015). Each review included studies that employed polysomnography and studies that employed actigraphy. Whilst Elrod and Hood (2015) and Díaz-Román et al. (2018) included many of the same papers, they used substantially different approaches to analysis. Elrod and Hood (2015) reviewed studies using polysomnography (in six cases), actigraphy (in four cases) and one study in which both were employed. Elrod and Hood (2015) analysed results of actigraphy and polysomnography in a single analysis. Across the 11 studies, there were significant differences between children with and without ASD on Total Sleep Time (on average 32.8 min per day shorter in ASD), Sleep Latency (10.9 min 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 & Schreck, 2009).

Díaz-Román et al. (2018) reviewed studies using polysomnography (in eight cases), actigraphy (in six cases) and one study in which both were employed. Plysomnography and actigraphy data were analysed separately. For polysomnography variables, children with ASD experienced shorter Total Sleep Time, longer Sleep Onset Latency, less Stage 1 sleep, less Rapid Eye Movement (REM) sleep, lower Sleep Efficiency, and earlier Wake Time. There was no difference in Stage 2 sleep, Slow Wave sleep or REM latency. For actigraphy, children with ASD experienced a longer Sleep Onset Latency, but no difference on measures of Sleep Duration or Efficiency.

In sum, current evidence from objective measures comparing sleep in children with ASD to their peers is equivocal. Polysomnography studies show clear and consistent differences, but employ methodologies that may have a significant impact on the sleep of children with ASD. Studies employing actigraphy, which is less invasive and includes longer testing periods, have shown less clear differences. The clearest evidence is for differences in Onset Latency, with some evidence for difference in Sleep Efficiency and little evidence for difference in Total Sleep Time. It is not clear whether these differences are also reflected in those children with an identified sleep problem.

1.4. Rationale

A growing body of research has examined the prevalence, severity, nature and cause of sleep problems in children with ASD. These studies have continually identified that sleep problems are more prevalent in children with ASD. However, relatively few of these studies have measured sleep objectively in comparison to a control group of children without ASD. The most regular method for this has been polysomnography, which may be hard to tolerate for some children with ASD and elicit anxiety. Studies employing actigraphy have been fewer and shown less robust differences between children with and without ASD. Further, more targeted research using actigraphy is required to understand sleep problems in children with ASD better.

The current study used actigraphy to compare sleep in children with and without ASD. 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). Parent diaries were also undertaken to compare parent reports of sleep in the two groups and to evaluate their accuracy using actigraphy. Sleep questionnaires investigated parent reports of specific problems with sleep. All children with ASD were tested using 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.

1.5. 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 objectively 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:

I Similarities and differences between actigraphy measures and parent reports.
II Individual differences in sleep quantity and quality using correlations.

2. Method

2.1. Participants

Sixteen children (see Table 1) with Autism Spectrum Disorders (mean age = 9.8 years; 63% male; average performance IQ = 100.7; average adaptive behaviour standard score = 77.4) were matched on age ($t(30) = .04$, $p = .83$, $d = .02$) and gender to 16 typically developing children. Diagnosis was corroborated through administration of the Autism Diagnostic Observation Schedule – second edition (ADOS-2; Lord et al., 2015, see below). Post-hoc comparisons showed no differences between the two groups on maternal education ($\chi2(3) = 6.95$, $p = .07$, $V = .481$) or family income grouping ($\chi2(6) = 10.10$, $p = .76$, $V = .40$). All families of children with ASD on a variety of databases[1] 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 5–15 years. Three more children were recruited to the original sample, but were excluded from this study. Two of these children were excluded because sleep diaries were

[1] 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.

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

Participant	Age (Years)	Gender	Performance IQ[a]	Adaptive Behaviour[b]
1	12	Female	112	68
2	7	Female	95	89
3	8	Male	131	75
4	13	Male	89	76
5	13	Male	94	79
6	11	Female	111	78
7	8	Male	110	80
8	12	Male	112	74
9	10	Male	70	62
10	9	Male	76	68
11	10	Male	77	66
12	7	Female	112	126
13	11	Female	127	68
14	10	Male	109	97
15	5	Male	> 103	65
16	10	Female	84	67

[a] Score on the Wechsler Abbreviated Scale of Intelligence (Wechsler, 1999).
[b] Standard Score on the Vineland Adaptive Behavior Scales (Sparrow et al., 2005).

Table 2
Group-level participant characteristics.

	ASD Group, Mean (Range)	TD Group, Mean (Range)	*t*	*p*	*d*
Age (Years)	9.8 (5-13)	9.5 (5-14)	.77	.96	0.02
Gender (%Male)	62.5%	62.5%			
Social Communication Questionnaire	27.62 (7.39)	3.29 (3.34)	11.17	< .001	8.78
Performance IQ	100.6 (70-131)				
VABS Adaptive Level	77.4 (62-126)				

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 thus 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 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 and hemiplegia). All parents of typically developing children selected scored below the ASD cut-off of 15 on the Social Communication Questionnaire (Rutter, Bailey, & Lord, 2003; indicative that none of them were likely to have ASD). Children with ASD scored significantly higher on the SCQ (see Table 2)

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)[2] .

2.2. 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 objectively 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.

2.2.1. Assessment of ASD, intellectual and adaptive functioning

Autism/ Autism Spectrum Disorder diagnoses were corroborated by completion of the ADOS-2 (Lord et al., 2015). All children 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 – 2nd edition (Sparrow, Cicchetti, & Balla, 2005) was used to measure adaptive behaviour and functioning.

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

2.2.2. Actigraphy

Each child wore an Actiwatch 2 (Phillips Respironics) on their non-dominant wrist for a continuous period of seven to eight days, in line with guidance on obtaining reliable measures of sleep through actigraphy (Acebo et al., 1999). 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 of sleep interval was identified by Actiware software and was corroborated against parent report in the sleep diary. The end of the sleep interval was identified by the end of 10 min 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 if 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).

2.2.3. 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 × (Total Sleep Time / Time in Bed).

2.2.4. Questionnaires

Parents completed a pack of questionnaires within a week of finishing the objective 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 to 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 to correlate with objective measurements of sleep.

2.2.4.1. Modified Simonds and Parraga sleep qestionnaire (Simonds & Parraga, 1982; Wiggs & Stores, 1996). The Simonds and Parraga Sleep Questionnaire is a broad ranging parent-report measure of sleep in children (Simonds & Parraga, 1982). The modified version was adapted for children with developmental disabilities (Wiggs & Stores, 1996). 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, Spirito, & McGuinn, 2000), $r = .70$. 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. The test-retest reliabilities of the subscales of the MSPSQ are 0.83–1 (Wiggs & Stores, 1996)

2.2.4.2. Family inventory of sleep habits (Malow, McGrew, Henderson, & Stone, 2006). The Family Inventory of Sleep Habits (FISH; Malow et al., 2006) 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. The FISH shows good test-retest reliability in children with ASD ($r = .82$) over three months and negative correlations with measures of sleep problems from the CSHQ (Malow et al., 2009).

2.2.4.3. Modified paediatric Epworth sleepiness scale (Williams, Scheimann, Sutton, Hayslett, & Glaze, 2008). The Epworth Sleepiness Scale (Johns, 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). The adult version of the scale demonstrates a good level of internal consistency ($\alpha = .88$; Johns, 1992) and reliable test-retest figures over a period of months ($r = .82$: Johns, 1992).

2.2.4.4. Social communication questionnaire (SCQ; Rutter et al., 2003). The Social Communication Questionnaire asks parents to report whether or not the child demonstrates a range of different social behaviours. It is often used as a quick scale parent report measure of ASD traits. The SCQ has been shown to be a good predictor of children's likelihood of having ASD ($AUC = .90$, Charman et al., 2007) and has good agreement with the Autism Diagnostic Interview- Revised (Rutter et al., 2003).

2.3. Data analysis

2.3.1. 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. Further to this, Bayesian independent samples *t*-tests were undertaken. This approach clarifies the support for null vs. alternative hypotheses. Here, the analysis strategy was to consider these when the initial *t*-test returned a non-significant result. The appropriate conclusion in these cases is that it cannot be concluded that the ASD and non-ASD groups significantly differed. Such conclusions have limitations. They rely on an arbitrary cut-off for concluding which data are allotted importance. Further, they provide no evidence as to whether the data support the null hypothesis or not. For clinical purposes this is of crucial importance. Knowing the degree to which data support commonality between two groups is just as important as knowing the degree to which it highlights group differences. Bayesian statistics provide such testing. The Bayes Factor represents the ratio of the probability of two hypotheses being true, given the data. For the current purposes, these hypotheses are that a) the two groups do differ and b) the two groups do not differ for a given variable.

For each of the t-tests undertaken, we provide a Bayes Factor - BF_{01}. This is a quantification of the support for the null hypothesis over the alternative hypothesis. A $BF_{01} = 1$ suggests that null and alternative hypotheses are equally consistent with the data. $BF_{01} > 1$ provides more support for the null hypothesis. In line with the underlying assumptions of Bayesian approaches, there are no "cut-offs" for "significant" differences. A widely used rule of thumb, following Jeffreys (1961) considers:

- BF_{01} = 1–3 as "anecdotal evidence" in favour of the null hypothesis
- BF_{01} = 3–10 as "moderate evidence" in favour of the null hypothesis
- BF_{01} = 10–30 as "strong evidence" in favour of the null hypothesis
- BF_{01} = 30–100 as "very strong evidence" in favour of the null hypothesis
- $BF_{01} > 100$ as "extreme evidence" in favour of the null hypothesis

Reciprocally, $BF_{01} < 1$ provides more support for the alternative hypothesis, such that

- BF_{01} = 1–1/3 as "anecdotal evidence" in favour of the alternative hypothesis
- BF_{01} = 1/10–1/3 as "moderate evidence" in favour of the alternative hypothesis
- BF_{01} = 1/30–1/10 as "strong evidence" in favour of the alternative hypothesis
- BF_{01} = 1/100–1/30 as "very strong evidence" in favour of the alternative hypothesis
- $BF_{01} < 1/100$ as "extreme evidence" in favour of the alternative hypothesis

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.

3. 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 analysed by using data from actigraphy. 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.

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

	ASD Mean (SD)	TD Mean (SD)	*t*	*p*	*d*	*Bayes Factor*
Bed Time (hh:mm)	20:46 (1:01)	21:26 (1:05)	1.75	.09	.62	1.10
Get-up Time (hh:mm)	06:46 (0:59)	07:15 (0:35)	1.70	.10	.60	1.18
Time in Bed (hh:mm)	09:59 (0:46)	09:49 (1:04)	.51	.62	.18	3.51
Total Sleep Time (hh:mm)	08:05 (0:39)	08:12 (0:45)	.50	.62	.18	3.52
Onset Latency (hh:mm)	00:38 (0:32)	00:29 (0:26)	.91	.37	.32	2.74
Sleep Efficiency (%)	82.08 (4.17)	83.88 (5.68)	1.02	.32	.36	2.51
Wake After Sleep Onset (hh:mm)	0:52 (0:17)	00:48 (0:16)	.68	.50	.24	3.21

Table 4
Group mean scores and differences for measurements from sleep diaries. *$p < .05$.

	ASD Mean (SD)	TD Mean (SD)	*t*	*p*	*d*	*Bayes Factor*
Bed Time (hh:mm)	20:25 (0:58)	20:55 (0:58)	1.44	.16	.51	1.65
Lights Out (hh:mm)	20:51 (1:05)	21:21 (1:09)	1.26	.22	.45	2.00
Wake up time (hh:mm)	06:39 (0:47)	07:13 (0:46)	2.05	.05*	.73	.71
Time out of Bed (hh:mm)	06:52 (0:49)	07:31 (0:45)	2.30	.03*	.81	.47
Time to get to Sleep (hh:mm)	0:39 (0:29)	0:32 (0:19)	.81	.43	.28	2.97
Wake After Sleep Onset (hh:mm)	0:11 (0:17)	0:03 (0:03)	1.87	.08	.65	1.06
Time in Bed (hh:mm)	10:28 (0:51)	10:25 (0:49)	.16	.87	.06	3.87
Total Sleep Time (hh:mm)	9:03 (1:07)	9:16 (1:22)	.45	.65	.20	3.58
Sleep Efficiency (%)	86.5% (8%)	88.7% (10%)	.70	.49	.25	3.18

3.1. 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.

In all cases, the Bayes factor suggests that these results are more consistent with the null hypothesis (that there is no difference between groups), than the alternative hypothesis (that the sleep of children with ASD is significantly different from their typically developing peers). Jeffreys (1961) suggests Bayes factor > 3 as substantial evidence in favour of the null hypothesis (Raftery, 1995; uses the same value to denote "positive" evidence). By these criteria, evidence for no difference between the groups is of particular note for: Total Sleep Time, Time in Bed, and Wake After Sleep Onset. For Bed Time, Get-up Time, Onset Latency, and Sleep Efficiency, the Bayes factors suggest that evidence in favour of the null hypothesis is weaker.

3.2. 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 4). There were trends for the children with ASD being reported to wake up and get out of bed earlier than the TD children.

Interpretation of the Bayes factors suggests that there is substantial evidence in favour of the null hypothesis for total sleep time, sleep efficiency, and time in bed. For bed time, lights out, time to get to sleep, and wake after sleep onset, evidence in favour of the null hypothesis was weaker.

3.3. 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 the MSPSQ (see Table 5). There was no evidence of group differences in sleep hygiene or day-time sleepiness (as measured by the FISH and the MPESS).

Interpretation of the Bayes factors suggest that there is substantial evidence in favour of the null hypothesis for the Epworth Sleepiness Scale, but weaker evidence for the Family Inventory of Sleep Habits.

3.4. 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 compared to only five of 16 typically developing children (31%). This represented a significant difference between groups ($X^2(1) = 9.02$, $p = .004$, $\varphi = .67$).

To investigate the likely sleep problems responsible for group differences on the MSPSQ, the subscales of the questionnaire were investigated. There were significant differences between the groups, such that the children in the ASD group scored higher than their

Table 5
Group mean scores and differences for questionnaires. **$p < .01$.

Variable	ASD Mean (SD)	TD Mean (SD)	*t*	*p*	*d*	*Bayes Factor*
Modified Simonds and Parraga Sleep Questionnaire	73.05 (15.16)	53.29 (13.59)	3.77	.001**	1.37	.024
Family Inventory of Sleep Habits	50.57 (11.06)	46.60 (5.02)	1.26	.22	.46	1.94
Modified Paediatric Epworth Sleepiness Scale	2.21 (3.02)	2.50 (1.97)	.31	.76	.11	3.65

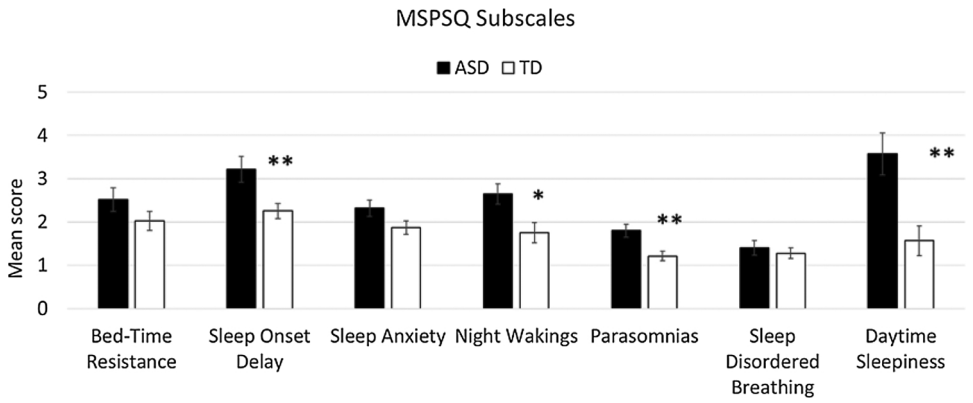

Fig. 1. Comparisons between groups on subscales of the Modified Simonds and Parraga Sleep Questionnaire. Error bars indicated standard error of the mean. $^{**}p < .01$, $^{*}p < .05$.

counterparts in the TD group on Sleep Onset Delay ($t(28) = 2.89$, $p = .007$, $d = 1.04$), Parasomnias ($t(28) = 3.22$, $p = .003$, $d = 1.17$), and Day-Time Sleepiness ($t(28) = 3.44$, $p = .002$, $d = 1.25$). There was also a trend for a difference on Night Waking ($t(28) = 2.68$, $p = .012$, $d = .98$, see Fig. 1). Wiggs and Stores (2004) suggest that any score above 4 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 ($t(28) \leq 1.87$, $p \geq .07$).

3.5. 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 (see Table 6). There were significant positive correlations between actigraphy data and diary reported bed-times, get-up times, times in bed, and total sleep times. 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 those 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 objective measurement reflected parents overestimating their children's sleep time.

3.6. 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 7). 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 in the ASD group. These correlations were large in magnitude ($r \geq 0.5$) and in the developmentally expected direction.

Table 6
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	*r*	*p*	*t*	*p*	*d*
Bed Time	Bed Time	.94	< .001**	3.98	.001**	.36
Get up time	Wake Time	.92	< .001**	1.05	.31	.12
Time in Bed	Time in Bed	.81	< .001**	3.74	.002**	.59
Total Sleep Time	Total Sleep Time	.68	.003**	4.74	< .001**	1.07
Onset Latency	Time to get to sleep	.50	.048*	.16	.87	.04
Sleep Efficiency	Sleep Efficiency	.15	.57	2.05	.06	.68
Wake After Sleep Onset	Wake After Sleep Onset	−.03	.90	6.80	< .001**	2.44

Table 7
Correlation between dependent variable questionnaires and sleep outcome measures (from actigraphy) in the ASD group *p < .05.

Measure	ASD		TD	
	Correlation with Total Sleep Time, ρ *(p)*	Correlation with Sleep Efficiency, ρ *(p)*	Correlation with Total Sleep Time, ρ *(p)*	Correlation with Sleep Efficiency, ρ *(p)*
Age	−.50 (.05*)	−.31 (.25)	−.32 (.22)	−.32 (.22)
Performance IQ	.07 (.80)	−.23 (.42)	N/A	N/A
Adaptive Behaviour (VABS)	.15 (.59)	−.54 (.03*)	N/A	N/A
Sleep Problems (MSPSQ)	.28 (.33)	−.03 (.91)	.29 (.27)	.13 (.62)
Sleepiness (PESS)	−.05 (.88)	−.11 (.72)	.25 (.36)	−.10 (.71)
Social Communication (SCQ)	.09 (.78)	.06 (.84)	.16 (.59)	.09 (.75)
Family Sleep Habits (FISH)	.22 (.46)	.23 (.42)	−.11 (.69)	−.26 (.35)

4. Discussion and implications

Sleep habits in children with ASD and a parentally-reported sleep problem were compared to sleep habits of children without ASD using a comprehensive range of measures. The most objective measurements of sleep, through actigraphy, found no significant differences between the two groups. Parent reports correlated well with objective measurements, other than for wakings. No significant group differences were found in parent diaries either. However, there was evidence of group differences in sleep on questionnaire measures.

4.1. Measuring sleep objectively through actigraphy

Consistent and reliable reports have concluded that sleep problems are more common in children with ASD, than they are in the general population; 44–83% of children with ASD experience sleep problems, while only 9–50% of children do more generally (Richdale & Schreck, 2009). It is therefore surprising that measurements of children's sleep taken through actigraphy were not significantly different for children with and without ASD. Such a conclusion is even starker 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 sleep duration (as measured through total sleep time) and quality of sleep (as measured through sleep onset, wake after sleep onset, and sleep efficiency). These findings are not entirely inconsistent with previous findings from actigraphy studies of children with ASD. In their recent meta-analysis, Díaz-Román et al. (2018) found no consistent differences between children with and without ASD for sleep duration. Elrod and Hood's (2015) meta-analysis reported similar findings for those children with ASD and no intellectual disability. The findings here are consistent with those results. Our follow-up Bayesian analysis is particularly relevant as it suggested that for sleep duration, these data provide substantial evidence in favour of the null hypothesis. Unlike for sleep duration, significant group-level differences between children with and without ASD have been found in sleep efficiency (Elrod & Hood, 2015) and sleep latency (Díaz-Román et al., 2018; Elrod & Hood, 2015). Here these were not observed. The follow-up Bayesian analysis of our data in relation to this was less decisive. These analyses suggested that the data would be twice as likely to be observed under the null hypothesis. This evidence is relatively weak in determining between null and alternative hypotheses.

The overall pattern of our data are broadly consistent with comparable studies. Baker et al. (2013) found no difference between sleep duration of high functioning adolescents with ASD and their peers, but did find marginally poorer sleep efficiency and sleep latency in the ASD group. Similarly Allik et al. (2006) found no difference between sleep duration in children with ASD and no comorbid intellectual disability and their peers. However, Allik et al. (2006) found poorer efficiency and longer latency. In sum, our results are consistent with the emerging literature on actigraphy studies comparing sleep in children with ASD and no comorbid intellectual disability to their peers. There is little evidence to support shorter differences in overall sleep time, but equivocal evidence for marginally longer latencies and poorer efficiency.

4.2. Parentally reported diaries

Parental reports of the sleep of their children with ASD correlated well with measures gained through actigraphy. This is further evidence that sleep diaries and actigraphy converge in estimating individual differences in sleep (Sadeh, 2011). Veatch et al. (2016) demonstrated this by finding commonality between actigraphy and parent-report measures in identifying specific areas of sleep difficulty in children with ASD. Although in the current study, the measures correlated, they were also significantly different, suggesting there were systematic differences in objective and subjective measures of sleep. Comparisons between the two measures showed that parents 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. Whilst objective 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), the current study provides evidence for the opposite. This stands as further evidence for the additional benefit of objective measures for research and clinical practice. Here, we suggest that by using diaries alone, researchers and clinicians may underestimate sleep difficulties. 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. Bayesian follow-up analyses suggested that for sleep duration and efficiency, the data provided substantial support for the null hypothesis.

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' reports of their child's sleep problem were not due to an inaccurate understanding of their child's sleep. One explanation is that parental experiences of sleep problems included difficulties with sleep other than duration of sleeping and waking, such as those identified by questionnaires.

4.3. 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, children with ASD scored significantly higher on the MSPSQ (Wiggs & Stores, 1996). This questionnaire assesses sleep and night-time behaviour more broadly than the actigraphy and diary measures. The MSPSQ (Wiggs & Stores, 1996) includes questions on bed-time resistance, day-time sleepiness, and parasomnias, as well as questions on sleep latency and night-time waking.

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. The MSPSQ items associated with day-time sleepiness report on drowsiness, but also increased activity. Over-activity is a common outcome of fatigue following poor sleep. Sleep disturbances are common in children with attention-deficit hyper-activity disorder (Ivanenko & Johnson, 2008) and are associated with challenging behaviour in ASD (Cohen, Conduit, Lockley, Rajaratnam, & Cornish, 2014).

Differences on the parasomnias subscale were informative. 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 (Ming, Sun, Nachajon, Brimacombe, & Walters, 2009; Williams et al., 2008). 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 confusional 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.

It was perhaps surprising that children with ASD did not score higher on the "bedtime resistance" subscale of the MSPSQ. These problems have been reported as particularly prevalent in children with ASD. For instance, Liu, Hubbard, Fabes, and Adam (2006) reported that 54% of children with ASD experience daily problems with bedtime resistance. One possibility is that this would have been a more significant problem had younger children been tested. Goldman, Richdale, Clemons, and Malow (2012) found that problems with bedtime resistance decreased with age in children with ASD. Another possibility is this related to the generally high levels of sleep hygiene (as evidenced by the FISH) in the ASD group. Bedtime resistance has been shown to correlate with poor sleep hygiene (Malow et al., 2009). That this group did not differ from their peers for bedtime resistance may go some way to explaining the lack of differences on objective and subjective measures of sleep time. A question for future research is whether children with ASD may not experience quantifiable differences in their sleep time if their parents can minimise bedtime resistance. Acceptance of this hypothesis has clear implications for sleep intervention,

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.

4.4. Limitations and future directions

Although an objective 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 Rapid Eye Movement 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.

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 and ability range (though all were in the normal range on a standardized measure of performance IQ). The small sample size limits power. We identified a number of significant differences, and the Bayesian analysis provided substantial evidence that a number of the null results were meaningful. On the other hand, the data provided only weak evidence to determine whether the data favoured the null or alternative hypothesis for some comparisons. This is notable in data on sleep efficiency and onset latency. The age range tested spans expected developmental changes in sleep. The case-control comparison goes some way to control for this, but it is likely that high within group variability was apparent. Equally, a sleep study may not attract a random sample of

typically developing children. Sleep efficiency in the typically developing group was lower than would typically be expected (84%). This suggests the study may have attracted some families whose typically developing children experience sleep difficulties. Further the typically developing group showed no increase in their sleep time with age, against developmental expectations. Concluding strongly about this should be tempered by the relatively small sample size for detecting correlations of this nature.

Equally, specificity of interpretation is more difficult across a more heterogeneous group. That the 16 children were drawn from only 10 families, with multiple siblings taking part, also provides a limitation. This has the opposite problem of reducing within group variance. All children were recruited on the basis of their parents reporting them to have a sleep problem. This was to follow the explicit aim of understanding sleep in children with ASD and sleep problems. It is acknowledged that this is different from understanding sleep in children with ASD more generally.

A further limitation is reflected in the testing of IQ and adaptive behaviour. The WASI is a brief measure of intellectual functioning, with only four subscales. Restricting to the perceptual reasoning composite meant that only two subscales were used to calculate the IQ reported. This was done as the verbal comprehension subscales often return particularly low scores in children with ASD (Mayes & Calhoun, 2003). The measures used relatively outdated norms for the WASI (1999) and the VABS-2 (2005). Scores on IQ tests have risen over time, meaning using old norms may result in over-estimation of a child's IQ in relation to their peers. Additionally to this, only the ASD group completed the ADOS-2, VABS-2, and WASI. Reports from parents of children in the control group suggested low levels of difficulty with social communication and no intellectual disability. However, this would have been more accurately confirmed by use of the direct/ observational measures used with the ASD group.

A decision was made to include children with ASD whose parents reported they had a sleep problem. The aim was to reflect those children with ASD seeking support for sleep problems. This meant children taking medication for sleep problems were included. This has the negative consequence of potentially underestimating the underlying sleep difficulties of those five children who were on medication at the time of the study. Unmedicated, their sleep may have been worse. It is worth noting that removing these children from analyses did not alter significance of results, suggesting their sleep profiles were similar to those of unmedicated peers with ASD. On the other hand, these may have been the children who initially had the very worst problems. With this in mind, the data reflect the sleep of children with ASD and parent-reported sleep problems in the community, not the expected sleep problems for those who have received no treatment.

4.5. Conclusions

Sleep problems in children with ASD have been well-documented and are cited as among the most common comorbid conditions for this group (Ming et al., 2007). However, there have been very few studies measuring sleep using actigraphy that have compared sleep in children with and without ASD. In our study, actigraphy was used to add to that literature. In support of a recent meta-analysis (Díaz-Román et al., 2018), there was no evidence that children with ASD 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 did not overestimate waking periods nor underestimated actual sleep for their children. Questionnaire data suggested that sleep problems in children with ASDs may have sleep problems that are not best captured by sleep time data.

Declaration of Competing Interest

None of the authors have any conflicts to report in relation to this work.

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