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# Cross syndrome comparison of sleep problems in children with Down syndrome and Williams syndrome

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ABSTRACT

Based on previous findings of frequent sleep problems in children with Down syndrome (DS) and Williams syndrome (WS), the present study aimed to expand our knowledge by using parent report and actigraphy to define sleep problems more precisely in these groups. Twenty-two school-aged children with DS, 24 with WS and 52 typically developing (TD) children took part in the study. Each child wore an actiwatch for a minimum of four nights and parents completed the Children's Sleep Habits Questionnaire (CSHQ). Sleep problems were common in both developmental disorders. Children with DS had the greatest sleep disruption, with frequent and longer night wakings as well as restlessness. Parents reported symptoms of sleep-disordered breathing and a range of other problems including grinding teeth, bedtime resistance and sleep anxiety. Children with WS had problems initiating sleep and parents also reported bed-wetting and body pain. Despite these problems, the mean actual sleep time, as measured by actigraphy, did not differ between the three groups. CSHQ reports were in agreement with actigraphy for children's sleep duration, but this was not the case for sleep latency, restlessness and the night wakings variables. Sleep problems in DS and WS are common and appear to be syndrome-specific. Due to the inaccuracy of parent report, it is recommended that children at risk undergo objective measures of sleep assessment.



## 1. Introduction

Sleep problems in TD children are common, with around one third experiencing some kind of sleep disturbance (Mindell & Owens, 2003; Owens, Spirito, McGuinn, & Nobile, 2000; Pegg, 2006). These range from behaviourally-based problems such as behavioural insomnia, to physiological problems such as sleep disordered breathing (SDB) and periodic limb movement disorder (PLMD). Parasomnias such as nocturnal enuresis (bed-wetting), somnambulism (sleep walking), bruxism (grinding teeth) or sleep terrors are also common but are generally outgrown by mid childhood.

Sleep is an important aspect of development and poor quality sleep has a detrimental effect on physical, cognitive and social functioning. Poor quality of sleep may manifest in behaviours such as daytime sleepiness, irritability, hyperactivity and impulsivity and have a negative effect on school performance (see Fallone, Owens, & Deane, 2000 for a review). Sleep

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problems can exert great pressure and distress on the family but are often amenable to treatment, which can improve the child's and family's quality of life (Merrell & Shott, 2007; Wood & Sacks, 2004).

It is now well established that sleep problems are also common in individuals with developmental disorders, yet there is relatively little information available characterising sleep patterns in these groups. This is perhaps surprising when considering the high frequency of sleep problems in such children and the importance of good sleep for healthy physiological and psychological development. For these reasons, reliable and valid methods for studying sleep in atypically developing children are crucial. The gold standard technique for the diagnoses of many sleep disorders is polysomnography (PSG): a detailed assessment in a sleep laboratory of physiological and neural activity during sleep. While this is a key diagnostic tool for sleep disorders that alter neurophysiology, such as PLMD, or respiratory function, such as in SDB, it only provides a single time point assessment that fails to capture dimensions of sleep associated with behaviour and usual sleep environment (Stores,Wiggs, & Campling, 1998). Furthermore, it is not always appropriate for children with developmental disorders who may become distressed at sleeping in a strange environment.

Relatively recently, actigraphy (movement monitoring) has emerged as a reliable and valid alternative which can be used to continuously assess activity levels in a home setting over a prolonged period of time. It yields more than 80% agreement with PSG in the prediction of wake and sleep for typical groups (Sadeh, Hauri, Kripke, & Lavie, 1995), but in comparison is inexpensive, non-intrusive, and less time consuming to analyse. In addition, it is more likely to be tolerated by children with developmental disorders. Its use can be supported by the use of questionnaires completed by parents to provide rich information on sleep habits and characteristics.

The aim of the current study is to use actigraphy supported by parent report to investigate common and syndrome-specific sleep problems in children with Down syndrome (DS) and Williams syndrome (WS) compared to a typically developing (TD) control group.

### 1.1. Down syndrome

DS is the most common sporadic genetic developmental disorder (1/700 live births) usually associated with the presence of three copies of chromosome 21 (trisomy 21). It is the leading genetic cause of intellectual disability, yielding an average IQ of around 50 points, but with wide individual variability (Roizen & Patterson, 2003). Individuals with DS often experience sleep disturbances (Carter, McCaughey, Annaz, & Hill, 2009; Cotton & Richdale, 2006). The most common of these is obstructive sleep apnoea syndrome (OSAS), a condition where the upper airway occludes during sleep, causing both disruption of sleep and intermittent hypoxia. Data from prospective community studies (Dyken, Lin-Dyken, Poulton, Zimmerman, & Sedars, 2003; Shott et al., 2006) and from children referred for clinical examination of SDB (Marcus, Keens, Bautista, Von Pechmann, & Ward, 1991) suggest that OSAS affects around two thirds of children with DS and is likely to be attributed to physical features associated with DS, such as craniofacial and upper airway abnormalities, obesity, tonsil and adenoid encroachment, and generalised hypotonia (Churchill, Kieckhefer, Landis, & Ward, 2011). Problems with settling, sleep maintenance and early morning waking have also been described from parent report studies (Breslin, Edgin, Bootzin, Goodwin, & Nadel, 2011; Cotton & Richdale, 2006; Stores, Stores, Fellows, & Buckley, 1998).

Much research assessing sleep in children with DS relies on parent reports, which may not always be as comprehensive and accurate as using objective methods. For instance, Shott et al. (2006) conducted an overnight PSG study with 56 young children with DS. They found evidence of SDB in 80% of their sample but when compared to parent report, it emerged that parents had both over- and under-reported problems, with only 23% being accurate. The high incidence of OSAS and the inaccuracy of parent reports together underline the importance of objective methods for studying sleep in individuals with DS.

### 1.2. Williams syndrome

WS is a rare neurodevelopmental disorder caused by a deletion of some 28 genes on the long arm of one copy of chromosome 7 at q11.23 (Donnai and Karmiloff-smith, 2000). Individuals with WS are inclined to be overly sociable and their performance is relatively proficient on language tasks, despite having an average IQ of 56 (range: 50–70) (Bellugi, Wang, & Jernigan, 1994; Mervis et al., 2000).

Similarly to the DS population, much previous data on sleep in WS has been acquired solely from questionnaire studies. These reported settling problems and night waking (Udwin, Yale, & Martin, 1987), as well as bed-wetting, getting up for the bathroom, and sleep anxiety (Sarimski, 1996). More recently, difficulties at bedtime, long sleep latencies, night waking as well as sleep anxiety have been reported (Annaz, Hill, Ashworth, Holley, & Karmiloff-Smith, 2011), and PLMD has found to be common (Arens et al., 1998; Goldman, Malow, Newman, Roof, & Dykens, 2009; Gombos, Bódizs, & Kovács, 2011). PSG has also demonstrated significant differences in sleep architecture, specifically decreased sleep efficiency, decreased REM sleep and increased slow wave sleep (Gombos et al., 2011; Mason et al., 2011). Although research thus far indicates significant sleep problems in WS, more in-depth research expanding this area is critical.

A cross-syndrome comparison of sleep problems in children with DS and WS will determine whether there are characteristic patterns specific to each disorder, or whether problems are common to both groups and could therefore be common to developmental disorders generally. Based on previous research, we expect to find significant sleep problems in children with DS and WS relative to TD children, but that there will be discrepancies between parent-report and actigraphy

findings. We also expect to find age-related changes to children's sleep, with older children having increasingly later bedtimes and shorter sleep duration.

## 2. Methods

### 2.1. Participants

Participants were 22 children with DS, 24 children with WS and a control group of 52 TD children. Fifty per cent of each group was female. Participant details are provided in Table 1. A one way ANOVA showed that there was no significant difference in age between the groups ($F(2,97) = .27$, $p = .77$). The majority of children were from middle socioeconomic background and were predominantly white. Families did not receive any incentive for taking part in the study.

Children were excluded from the study if they had co-morbid medical or psychiatric disorders likely to affect sleep such as epilepsy and poorly controlled asthma or eczema; if they were taking any hypnotic medication and if English was not their first language.

TD children were recruited through local primary schools. Parent of children with DS were contacted through mailing lists of local support groups, special needs schools and groups. TD and DS families were given written information stating that the study was investigating normal sleep patterns in children. They responded either to the school or directly to the researcher if they wished to take part. Sixty parents of TD children responded to recruitment information; of these, one had a medical condition, two did not have English as their first language, and two later declined to take part. Of the 40 DS families who responded, six lived too far away, two would not tolerate the actiwatch, four had comorbid disorders or medical conditions, and three later declined to take part. Of the remaining 55 TD children and 25 children with DS who met the inclusion criteria, participants were selected based on age and sex in order to create balanced groups.

Contact details for parents of children with WS were made available by the Williams Syndrome Foundation UK. Families of school-age children were contacted by telephone and later in writing. Of all families contacted only two declined to take part in the study.

All children in the DS group had previously been tested positively for full trisomy of chromosome 21. Children with WS had been diagnosed clinically and by fluorescence in situ hybridization analysis for the detection of hemizygosity at the elastin locus (7q11.22-11.23). Ethical approval was granted by the Institute of Education, University of London Research Ethics Committee and supported by Down Syndrome Education International and The Williams Syndrome Foundation, UK.

### 2.2. Procedure

Parents were contacted by telephone to arrange participation and families were visited at home where they provided written informed consent. All children were asked if they wished to take part and all gave their verbal assent.

#### 2.2.1. Actigraphy

Each child was given an Actiwatch Mini (CamNTech, Cambridge, UK) to objectively assess their sleep patterns. This was worn around the non-dominant wrist, as if wearing a watch, and children were asked to wear it continuously for 1 week, as is recommended by Acebo et al. (1999). Data were downloaded to a computer and were analysed using Sleep Analysis 7 at the default 'medium' sensitivity level (CamNTech, Cambridge, UK). The programme uses an algorithm to score each 1-min epoch as sleep or wake based on movement during that minute, as well as the two preceding and two successive minutes. Sleep start and sleep end were marked as the start and end respectively of a period of 10 or more minutes of immobility. The actigraphy variables of interest are: bed time (time the child is in bed with lights off), time in bed, sleep latency (time from bed time to sleep start), assumed sleep time (time from sleep onset to offset), actual sleep time (assumed sleep minus any periods of wake), night wakings (number of), wake after sleep onset, sleep efficiency (percentage of time spent asleep from sleep onset to offset), moving time (percentage of time spent moving from sleep onset to offset) and fragmentation (an indication of restlessness where a higher figure denotes increased restlessness).

In addition, parents completed a sleep diary recording their child's bed time, getting up time, and any daytime naps or night-time awakenings for the duration of the study. These diary parameters were used to support analyses of actigraphy data.

#### 2.2.2. Children's Sleep Habits Questionnaire

Parents completed the Children's Sleep Habits Questionnaire (CSHQ; Owens, Spirito, & McGuinn, 2000). This is a 33-item caregiver response questionnaire which screens for the occurrence of common sleep problem symptoms in school-aged

**Table 1**
Participant details.

| Group | Chronological age (mean (SD)) | Age range |
|---|---|---|
| TD ($n = 52$) | 9.23 (1.69) | 6.02–12.90 |
| DS ($n = 22$) | 9.42 (1.98) | 6.09–12.23 |
| WS ($n = 24$) | 9.55 (2.09) | 6.08–12.58 |

children. Parents indicate whether each listed characteristic occurs often (5–7 nights per week), sometimes (2–4 nights per week) or rarely (0–1 nights per week) for behaviours such as going to bed at the same time each night, sleepwalking, bruxism or snoring. The CSHQ yields scores on eight subscales: bedtime resistance, sleep onset delay, sleep duration, sleep anxiety, night waking, parasomnias, SDB and daytime sleepiness, as well as a total sleep disturbance score. Satisfactory test–retest reliability of CSHQ subscales has been reported for both normal and clinical populations (Owens et al., 2000a, b).

#### 2.2.3. Statistical analysis

Data were analysed using SPSS for Windows, Version 19 (SPSS Inc., Chicago, IL) and screened for outliers using Cook's distance and Levene's test to assess the assumption of homogeneity of variance. Outlying scores were individually removed, this was never more than two scores per variable. Firstly, Analysis of Variance (ANOVA) on the actigraphy and CSHQ variables for each group was carried out followed by inter-group comparisons. Post-hoc comparisons were made using the Bonferroni test. Secondly, Pearson's correlations were used for the comparisons of the sleep measures across the groups. Third, we computed Analysis of Covariance (ANCOVA) tests to compare developmental trajectories of the groups with chronological age as a covariate (Thomas et al., 2009). Separate analyses were conducted to avoid masking any effects found in single groups by the difference in variability between groups (e.g., Annaz et al., 2010).

## 3. Results

One child from the TD group refused to wear the actiwatch. All other children had four or more days and nights of actigraphy data and the majority (78%) had seven or more, as requested. The numbers of nights recorded for each group are presented in Table 2. One-way between-groups ANOVA showed no significant difference in compliance between the groups ($F(2, 94) = 1.72$, $p = .19$, $\eta p^2 = .04$). CSHQ data were missing for five TD children and for two children with DS as parents did not return the questionnaire.

### 3.1. Actigraphy

Each actigraphy variable was analysed and compared between the groups using one-way between-groups ANOVAs. Results showed that TD children went to bed significantly later than both other groups. Children with WS had the longest sleep latencies, whilst children with DS had the most disrupted sleep (see Table 3).

### 3.2. Children's Sleep Habits Questionnaire

TD and cross-syndrome comparisons of scores on the CSHQ were made using one-way between-groups ANOVAs. Relative to the TD group, children with DS had significantly elevated scores on all subscales except sleep onset delay and daytime sleepiness, whilst children with WS had significantly higher scores for sleep onset delay, sleep duration and night wakings (see Table 4).

### 3.3. Comparison of actigraphy and CSHQ measures

Where analogous variables existed, Pearson's product moment correlations were used to investigate the similarity between parent report and actigraphy. This was conducted for assumed sleep time, sleep onset latency, fragmentation/restlessness, number of night wakings, and night waking duration. The only variable that showed a significant positive relationship between the actigraphy and CSHQ scores was for assumed sleep time (TD: $r(46) = .51$, $p < .01$; DS: $r(20) = .65$, $p < .01$; WS: $r(24) = .43$, $p < .05$). Sleep latency was expected to correlate with parentally reported sleep onset delay. Results showed a trend towards agreement between parent report and actigraphy but this did not reach significance for any group (TD: $r(46) = .25$, $p = .09$; DS: $r(20) = .30$, $p = .20$; WS: $r(24) = .40$, $p = .05$). For restlessness, and number and duration of night wakings, no significant relationships between parent report and actigraphy emerged in any group. In fact, many of the correlations were negative, indicating that parents' knowledge of their children's sleep is limited.

### 3.4. Developmental association with actigraphic sleep parameters

ANCOVAs were used to investigate whether sleep parameters were associated with the chronological age of individuals. In the two developmental disorder groups, there was an expected general trend for a decrease in sleep time with increasing

**Table 2**
Number of nights recorded by actigraphy for each group.

| Group | Mean (SD) | Median | Range |
|---|---|---|---|
| TD ($n = 51$) | 6.57 (1.33) | 7 | 4–10 |
| DS ($n = 22$) | 7.14 (1.21) | 7 | 5–11 |
| WS ($n = 24$) | 6.75 (.85) | 7 | 5–8 |

**Table 3**
Group mean scores and group differences using ANOVA on selected actigraphy variables.

| | TD ($n = 51$) | DS ($n = 22$) | WS ($n = 24$) | $F$ | $p$ |
|---|---|---|---|---|---|
| Bed time (hh:mm) | **21:32 (00:39)** | 20:30 (00:37) | 20:52 (00:37) | 22.04 | <.001*,† |
| Time in bed (hh:mm) | 10:09 (00:34) | **10:36 (00:39)** | 10:15 (00:48) | 3.65 | .03* |
| Sleep latency (hh:mm) | 00:25 (00:11) | 00:23 (00:16) | **00:48 (00:37)** | 10.70 | .02†,‡ |
| Assumed sleep time (hh:mm) | 09:31 (00:33) | **10:06 (00:43)** | 09:18 (1:00) | 7.35 | .001*,‡ |
| Actual sleep time (hh:mm) | 08:22 (00:32) | 08:22 (00:50) | 08:16 (00:53) | .19 | .83 |
| Night wakings | 31.53 (7.74) | **39.50 (9.19)** | 27.41 (5.93) | 14.70 | <.001*,‡ |
| Wake after sleep onset (hh:mm) | 01:07 (00:22) | **01:43 (00:33)** | 1:02 (00:22) | 20.02 | <.001*,‡ |
| Sleep efficiency (%) | 87.86 (3.81) | **82.99 (5.45)** | 88.93 (3.38) | 3.67 | <.001*,‡ |
| Moving time (%) | 13.89 (3.20) | **20.47 (5.00)** | 13.81 (3.25) | 26.98 | <.001*,‡ |
| Fragmentation | 28.93 (7.09) | **41.76 (8.74)** | 30.76 (7.02) | 23.08 | <.001*,‡ |

Significant problems are highlighted in bold.
* Significant difference between TD and DS ($p < .05$).
† Significant difference between TD and WS ($p < .05$).
‡ Significant difference between DS and WS ($p < .05$).

age. This trend was significant for assumed sleep time in the WS group but not for the other two groups. There was a significant decrease with age for actual sleep time for the DS and WS groups but not for the TD group. Perhaps surprisingly, no such trend for later bedtime or earlier getting up time was shown except that in the TD group chronological age significantly explained 12% of the variance in bedtimes with older children going to bed later. See Table 5 and Figs. 1 and 2.

### 3.5. Parentally reported sleep problems

Some interesting data are lost through amalgamating scores to subscales, so individual items of the CSHQ were investigated in further detail to establish whether group differences were apparent. The results in Table 6 indicates the percentage of parents who reported sleep characteristics occurring sometimes and usually (two or more nights per week). ANOVAs were then used to establish whether the groups were significantly different on these variables.

## 4. Discussion

In line with the previous studies (e.g., Annaz et al., 2011; Carter et al., 2009), the current data show that both developmental disorder groups have sleep problems. This, however, is the first direct cross-syndrome comparison of sleep in atypically developing populations. Children in the DS group experienced greater problems with sleep than the WS and TD groups. In the DS group, sleep problems were reported for almost all subscales of the CSHQ as well as total CSHQ score. Atypically high scores on bedtime resistance, sleep anxiety and night wakings subscales are consistent with previous literature describing settling and sleep maintenance difficulties in DS (Stores et al., 1998a). Parents of children with DS also reported a higher incidence of SDB symptoms on the CSHQ. In addition, actigraphy data showed more fragmented sleep with increased movement and night wakings. These symptoms are suggestive of OSAS, which is known to be common in DS (Shott et al., 2006), but a limitation of the present study was that it did not make use of pulse oximetry to establish the presence of hypoxia. Mean score on the daytime sleepiness subscale of the CSHQ was not significantly elevated for children with DS, who scored only marginally higher than the other two groups. This is in contrast to most previous research which found daytime sleepiness to be a particular problem for individuals with DS. For example, Breslin et al. (2011) found that their sample of 35

**Table 4**
Group mean scores and group differences using ANOVA on CSHQ.

| Subscale (possible score range) | TD ($n = 47$) | DS ($n = 20$) | WS ($n = 24$) | $F$ | $p$ |
|---|---|---|---|---|---|
| Bedtime resistance (6–18) | 7.00 (1.67) | **8.55 (2.48)** | 7.21 (1.44) | 5.18 | .05* |
| Sleep onset delay (1–3) | 1.35 (.48) | 1.55 (.76) | **1.83 (.76)** | 4.47 | .03† |
| Sleep duration (3–9) | 3.70 (1.15) | **4.85 (1.81)** | **4.71 (1.73)** | 5.70 | .01*,† |
| Sleep anxiety (4–12) | 5.11 (1.28) | **6.25 (1.33)** | 5.08 (1.98) | 4.49 | .01*,‡ |
| Night wakings (3–9) | 3.38 (.61) | **6.35 (1.73)** | **5.25 (1.59)** | 45.21 | <.001*,†,‡ |
| Parasomnias (7–21) | 8.87 (1.45) | **10.40 (2.30)** | 9.29 (1.55) | 5.66 | .01* |
| Sleep disordered breathing (3–9) | 3.20 (.46) | **5.45 (1.64)** | 3.63 (1.10) | 34.95 | <.001*,‡ |
| Daytime sleepiness (8–24) | 11.67 (2.51) | 12.45 (2.83) | 11.75 (2.98) | .64 | .51 |
| Total score (33–99) | 42.67 (5.38) | **52.90 (7.57)** | 46.50 (7.55) | 18.23 | <.001*,‡ |

Significant problems are highlighted in bold.
* Significant difference between TD and DS ($p < .05$).
† Significant difference between TD and WS ($p < .05$).
‡ Significant difference between DS and WS ($p < .05$).

**Table 5**
ANCOVA results showing relationship between chronological age and selected actigraphy variables.

| Variable | | *F* | *p* | Effect size |
|---|---|---|---|---|
| Bed time | TD ($n = 51$) | 6.15 | .02 | .12 |
| | DS ($n = 22$) | 3.49 | .08 | .15 |
| | WS ($n = 24$) | 1.23 | .28 | .05 |
| Get up time | TD ($n = 51$) | .15 | .70 | .003 |
| | DS ($n = 22$) | .13 | .73 | .01 |
| | WS ($n = 24$) | 1.56 | .23 | .07 |
| Assumed sleep time | TD ($n = 49$) | 2.64 | .11 | .05 |
| | DS ($n = 22$) | 2.59 | .12 | .12 |
| | WS ($n = 24$) | 7.29 | .01 | .25 |
| Actual sleep time | TD ($n = 49$) | 1.61 | .21 | .03 |
| | DS ($n = 22$) | 5.05 | .04 | .20 |
| | WS ($n = 24$) | 6.88 | .02 | .24 |

children with DS aged 7–18 (mean age 12.65) had a significantly elevated mean score for the daytime sleepiness subscale and 60% routinely fell asleep whilst riding in the car or watching television. When individual items were investigated, the current study showed similar findings. Children with DS were almost three times more likely than TD children to appear sleepy during the day, particularly whilst riding in the car and watching television, but this did not significantly increase the mean subscale score. These symptoms could also be a manifestation of OSAS. Breslin et al. (2011) found significant sleep problems in almost all areas, with 85.7% of the DS sample having sleep disturbance scores in the clinical range. This is slightly lower than the 95% found in the present study, yet is probably accounted for by the slightly higher age range and their finding that some sleep problems (e.g., sleep anxiety) decline with age. This was not found in the present study, but may reflect the relatively narrow age range.

For the WS group, the greatest problem appeared to be long sleep latencies reported by parents and evidenced in the actigraphy recording, although there was also wide inter-individual variability. Mean sleep latency was 48 min, 23 min longer than the TD group and 25 min longer than the DS group. Although parents reported night wakings and short sleep duration to be a problem in the WS group, this was not reflected in the actigraphy data, and in fact the WS group had fewer night wakings than the TD group. This inconsistency may be due to a reduced ability in the WS group to self-soothe back to sleep, therefore demanding more attention from parents and leading to increased parental report of night waking, despite objective measures being comparable to the TD group. TD children learn to self-soothe in infancy, with around 60–70% of infants being able to self soothe by 12 months of age (Anders, Halpern, & Hua, 1992).

The present findings are not completely consistent with our previous data from 64 children with WS, where particular problems were found on the bedtime resistance, sleep onset delay, sleep anxiety, night waking and daytime sleepiness subscales (Annaz et al., 2011). Perhaps this disparity is due to the greater effect of a much larger sample. In concurrence with this study, we also found a high level of bedwetting in the WS group, which could be attributed to frequent urine infections and urinary system anomalies, known to be common in WS (Schulman, Zderic, & Kaplan, 1996).Parent report was not always in agreement with actigraphy data. Although parents were reasonably accurate at reporting how long children had slept, no other correlations were found between actigraphy and CSHQ. It was expected that parent report should be quite precise for sleep latency, and although positive correlations were found for all groups, none reached the significance level of .05. One reason for this may be that the CSHQ subscale for sleep onset delay consists of only one item (Child falls asleep within 20 min), yielding a score of 1 (rarely), 2 (sometimes) or 3 (usually). This is perhaps not detailed enough to correlate with the range of sleep latency times experienced by children. Parent report was also inconsistent with actigraphy data for restlessness and night wakings, possibly because without constant watch over the child, it would be impossible to know the

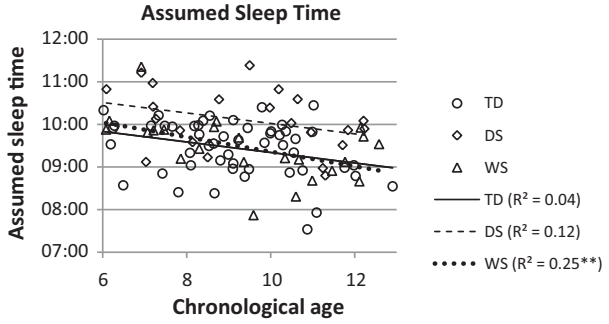


**Fig. 1.** Showing decrease in assumed sleep time with increasing chronological age (**$p < .01$).

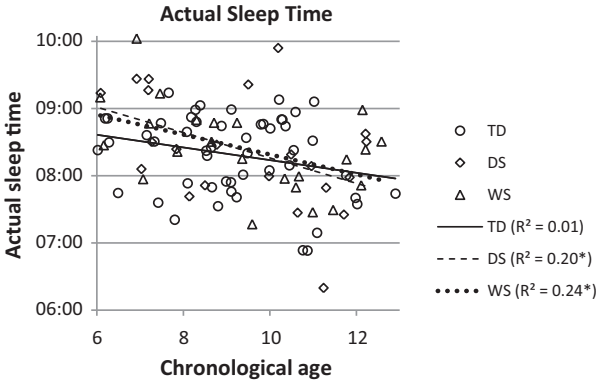


**Fig. 2.** Showing decrease in actual sleep time with increasing chronological age (*$p < .05$).

frequency of these behaviours. These findings support previous research suggesting that parents are not always aware of characteristics of their children's sleep (Shott et al., 2006; Wiggs, Montgomery, & Stores, 2005) and that whilst parents are reasonably good at reporting sleep start and finish times, they are less accurate in other areas (Lam, Mahone, Mason, & Scharf, 2011). Conversely, trends were observed for sleep latency, so perhaps with a larger sample size, greater power, or more precise questioning these figures would have been significant.

Despite its inaccuracy on these subjective issues, it was thought that although parent report might be more useful for uncovering specific information regarding sleep, for example parasomnias such as bed-wetting or grinding teeth, hence these and other items were investigated individually. This is particularly important for the parasomnias subscale, where items are unrelated so rich information is lost by amalgamating scores. This analysis revealed key differences between groups. Children with DS and WS were much more likely than typical children to move to another person's bed during the night. Bedwetting and complaining of body pain were particular problems for children with WS, whilst restlessness, bruxism, loud snoring and early waking were problems for the DS group. Children with DS were also more likely than other groups to wake in a negative mood, although the difference was not statistically significant. Almost half of the TD children were reported to talk in their sleep. Parent report is useful for gaining information on these parameters since most cannot be measured by actigraphy. In addition, a well-kept sleep diary is necessary for analysis of actigraphy data so parent report should still be valued. However, due to its inaccuracy for reporting subjective sleep characteristics, it should not be used as a substitute for objective measures, but ought to be used in conjunction. The present study benefits from using both objective and subjective techniques to support one another.

Although there are no established norms, Owens et al. (2000a, b) used the CSHQ with a sample of 494 school-aged children. For the TD group our findings are quite similar, except that our sample scored much higher on the Daytime Sleepiness subscale (11.67 compared to 9.63) and total sleep problems score (42.67 compared to 38.80). This may represent cultural sleep differences between our UK populations and the US sample on which the original data were based.

Actigraphy showed that the three groups were remarkably similar in the actual amount of sleep they achieved. The means for the three groups were within 6 min of one another, but standard deviations were much larger in the developmental

**Table 6**
Cross-syndrome comparison using ANOVA of the percentage of children expressing sleep characteristics on two or more nights per week.

| Variable | TD ($n = 47$) | DS ($n = 20$) | WS ($n = 24$) | $F$ | $p$ |
|---|---|---|---|---|---|
| Wets the bed | 6 | 30 | **45** | 7.56 | .01[†] |
| Restless sleep | 57 | **95** | 67 | 13.73 | <.001[*,‡] |
| Sleep talks | **47** | 30 | 17 | 3.82 | .02[‡] |
| Sleep walks | 6 | 10 | 4 | .30 | .74 |
| Moves to another bed | 15 | **60** | **50** | 12.95 | <.001[*,†] |
| Reports body pain | 11 | 15 | **33** | 3.10 | .15[†] |
| Grinds teeth | 23 | **45** | 8 | 3.33 | .04[‡] |
| Snores loudly | 19 | **70** | 29 | 11.98 | .001[*,‡] |
| Has frightening dreams | 17 | 25 | 30 | .60 | .55 |
| Wakes very early | 32 | **75** | 54 | 4.31 | .02[*] |
| Wakes in negative mood | 28 | 45 | 17 | 2.17 | .10 |
| Seems tired during day | 32 | **85** | 58 | 10.10 | <.001[*] |
| Complains about sleep problems | 15 | 15 | 17 | .29 | .75 |

Significant problems are highlighted in bold.
[*] Significant difference between TD and DS ($p < .05$).
[†] Significant difference between TD and WS ($p < .05$).
[‡] Significant difference between DS and WS ($p < .05$).

disorder groups, indicating greater variability. Despite the resemblance, the DS group had considerably more night wakings, wake after sleep onset, and lower sleep efficiency, therefore spending significantly longer in bed to achieve this sleep time.

The age range of the study is a developmental period associated in TD populations with a mean reduction in night sleep of 1.7 h (Iglowstein, Jenni, Molinari, & Largo, 2003). We would have expected this decline in sleep time to be apparent in the present study. In general there was a trend for children having less sleep with increasing age, which was significant in the DS and WS groups, although this was not associated with bedtime or getting up time. Children in the TD group went to bed later with increasing age, but this was not related to their total sleep time. These results suggest that children in all groups adjust their sleep time to suit their age-related needs, irrespective of bedtime.

### 4.1. Limitations

A limitation of the study is that although the Actiwatch Mini is assumed to be comparable to other validated devices, to our knowledge, it has not yet been validated against PSG or other actigraphs. Nor has it been validated for use in children with developmental disorders to determine whether it is the best method for objectively measuring sleep. Our use of a TD group gives a useful comparison in this case, so the data are valuable even if they cannot necessarily be compared to the wider literature. In addition, Meltzer and Westin (2011) compared two different scoring algorithms and, although they gave different results, the disparities were not clinically meaningful so data may still be compared across studies.

Participants in the TD and DS groups were self-selecting so parents may have been keener to take part if their child experienced sleep problems. An attempt was made to minimise this effect by stating in the recruitment material that the study was investigating normal sleep patterns, however, we cannot be certain that there was no response bias and that, therefore, sleep problems in these groups may be worse here than would normally be found in a random sample. For the WS group, we are more confident that the findings are generalizable to the WS population, since only two families declined to take part.

A further limitation is that demographic data were not collected, meaning that children could not be matched on these variables, and that those potentially affecting sleep could not be ruled out; for example, ethnicity, socioeconomic status, number of siblings, or parental age. Children were not selected based on these factors so it is hoped that they were representative of the populations from which they were drawn. Future studies would benefit from detailed demographic information in order to control for effects of these variables. Future work could also include pulse oximetry in order to assess whether sleep disruption could be attributed to OSAS symptoms, particularly in the DS group, where OSAS is known to be common.

### 4.2. Conclusion

This study effectively used actigraphy to study sleep patterns over the course of 1 week in children with DS and WS as well as a large control group. This approach offers considerable benefits in such children who are challenging to study in a laboratory environment. Much previous research has relied solely upon parent report, but we have demonstrated here that its inaccuracy necessitates the concurrent use of objective measures.

In conclusion, our study suggests that sleep problems are syndrome-specific. Children with DS experience more resistant behaviours at bedtime and sleep maintenance difficulties, the latter most likely due to SDB symptoms. Children with WS experience long sleep latencies, although are not reported to be resistant to settling. However, once asleep, their sleep quality is remarkably good although they do experience some parasomnias. The study extends previous work in WS, where research on sleep is limited, and provides a direct cross-syndrome comparison as well as a control group. The findings show that sleep problems are common and highlight the need for objective assessment in atypical groups.

## Financial disclosure and conflict of interest

The authors have indicated they have no financial relationships to this article to disclose. This manuscript has not been previously published and has not been submitted elsewhere.

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