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Links between sleep and daytime behaviour problems in children with Down syndrome

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Abstract

Background In the general population, sleep problems have an impact on daytime performance. Despite sleep problems being common among children with Down syndrome, the impact of sleep problems on daytime behaviours in school-age children with Down syndrome is an understudied topic. Our study examined the relationship between parent-reported and actigraphy-measured sleep duration and sleep quality with parent and teacher reports of daytime behaviour problems among school-age children with Down syndrome.

Method Thirty school-age children with Down syndrome wore an actigraph watch for a week at home at night. Their parent completed ratings of the child's sleep during that same week. Their parent and teacher completed a battery of measures to assess daytime behaviour.

Results Parent reports of restless sleep behaviours on the Children's Sleep Habits Questionnaire, but not actigraph-measured sleep efficiency, was predictive of parent and teacher behavioural concerns on the Nisonger Child Behaviour Rating Form and the Vanderbilt ADHD Rating Scales. Actigraph-measured sleep period and parent-reported sleep duration on the Children's Sleep Habits Questionnaire was predictive of daytime parent-reported inattention. Actigraph-measured sleep period was predictive of parent-reported hyperactivity/impulsivity.

Conclusion The study findings suggest that sleep problems have complex relationships to both parent-reported and teacher-reported daytime behaviour concerns in children with Down syndrome. These findings have implications for understanding the factors impacting behavioural concerns and their treatment in school-age children with Down syndrome.

Keywords behaviour, children, Down syndrome, sleep, trisomy 21

Children with Down syndrome demonstrate a variety of sleep problems, including obstructive sleep apnoea (OSA), sleep onset difficulties, frequent night awakenings and premature awakening (Marcus *et al.* 1991; Stebbens *et al.* 1991; Epstein *et al.* 1992; de Miguel-Diez *et al.* 2003; Carter *et al.* 2009; Churchill *et al.* 2014; Esbensen 2016; Maris *et al.* 2016). OSA affects 31–66% of individuals with Down syndrome (Stebbens *et al.* 1991; de Miguel-Diez *et al.* 2003), and behavioural sleep disturbances affect 52–69% of children with Down syndrome (Carter *et al.* 2009; Esbensen and Hoffman 2017).

In the general population, paediatric sleep problems are associated with deficits in neurobehavioural functioning including attention, impulse control, cognitive abilities (e.g. learning/memory) and behavioural regulation (Fallone *et al.* 2002; Steenari *et al.* 2003; Paavonen

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et al. 2009; Paavonen *et al.* 2009; Dewald *et al.* 2010; Beebe 2011) as well as adverse outcomes for parents and the family (Sadeh *et al.* 2000; Sadeh *et al.* 2002; Beebe 2006; Dahl 2006). Among children with intellectual and developmental disabilities, preliminary studies suggest that sleep problems may similarly be associated with maladaptive behaviours (Stores and Wiggs 2001). Parent-reported sleep concerns in children with autism spectrum disorders (ASD) demonstrate a positive correlation with parent reports of behaviour problems on general behaviour rating scales (Malow *et al.* 2006), and there is some evidence that improving sleep in children with ASD and severe insomnia results in improved behavioural functioning (Wright *et al.* 2011). Further, parent-reported sleep concerns in children with idiopathic intellectual disability also demonstrate a positive correlation with maladaptive behaviours (Quine 1991; Richdale *et al.* 2000; Didden *et al.* 2002; Rzepecka *et al.* 2011). Specifically, parent reports of sleep problems on the Children's Sleep Habits Questionnaire (CSHQ) are associated with parent reports of both anxiety and challenging behaviour (Rzepecka *et al.* 2011). Additionally, sleep problems in children with intellectual and developmental disabilities are related to parental stress and well-being, parenting behaviours (e.g. discipline, demonstrating affection) and parental sleep (Quine 1991, 1992; Richdale *et al.* 2000). While poorer attention has been speculated to be associated with sleep difficulties in children with intellectual and developmental disabilities (Harvey and Kennedy 2002), at least one study found that inattention was unrelated to sleep problems, as measured by actigraphy and pulse oximetry (Ashworth *et al.* 2015).

The relationship between sleep and daytime behaviour has particular relevance among children with Down syndrome, for two primary reasons. First, children with Down syndrome are at an elevated risk for daytime behaviour problems that are associated with sleep in other clinical populations. For example, children with Down syndrome often engage in noncompliant behaviours (McGuire and Chicoine 2006) and are characterised by symptoms of attention deficit hyperactivity disorder at rates two to three times higher than similarly aged children with intellectual and developmental disabilities (Dekker and Koot 2003; Froehlich *et al.* 2007; Ekstein *et al.* 2011). Second, both physiologically based sleep problems [e.g. OSA (Bull and Genetics 2011)] and behaviourally based sleep problems (e.g. bedtime resistance) have high prevalence in Down syndrome, with more concerns observed in children than adults (Esbensen 2016) and in pre-adolescents than adolescents (Carter *et al.* 2009). Individuals with Down syndrome also frequently demonstrate abnormal polysomnography with high apnoea hypopnoea index and sleep fragmentation (Levanon *et al.* 1999; Trois *et al.* 2009).

To date, very few studies have examined the relationship between sleep and daytime behaviour in children with Down syndrome. In studies among school-age children with Down syndrome, disturbed sleep was associated with both the child's daily functioning (irritability and overactivity) and parental well-being (Stores 1993; Stores *et al.* 1998). Parent-reported sleep concerns among children with Down syndrome have also been associated with worse adaptive functioning compared with typically developing children, particularly with respect to personal care, relationships, school, fitness and communication (Churchill *et al.* 2014). These studies have exclusively relied on parent reports of sleep and behavioural concerns, which can tend to inflate associations due to shared reporter variance, and with limited external validation of their measures. Using a small sample, actigraphy measures of total sleep time, activity level and snoring are correlated with several measures of inappropriate behaviour in children with Down syndrome (Stores and Stores 2014). These preliminary studies beg for additional research to confirm the relationship between sleep and behaviour in other contexts and to confirm the importance of evaluation and management of sleep problems in the clinical care for children with Down syndrome.

Psychometrically validated measures are available to assess sleep and maladaptive behaviour in children with Down syndrome. The CSHQ is commonly used to assess sleep in children with intellectual and developmental disabilities (Carter *et al.* 2009). The CSHQ has been demonstrated to be psychometrically sound for children with Down syndrome (Esbensen and Hoffman 2017). Children with Down syndrome demonstrate elevated concerns relative to typically developing peers for bedtime resistance, sleep onset delay, sleep anxiety, night waking, parasomnias, sleep disordered breathing and daytime sleepiness subscales of the CSHQ (Carter *et al.* 2009; Esbensen

and Hoffman 2017). However, parent reports of disordered breathing do not correspond with abnormal polysomnography findings (Shott *et al.* 2006). The Nisonger Child Behaviour Rating Form (NCBRF) is currently recommended for use in children with Down syndrome for assessing comorbid behaviours of concern (Edgin *et al.* 2010). Both the NCBRF and the Vanderbilt ADHD Rating Scales are considered appropriate for children with intellectual and developmental disabilities (Esbensen *et al.* 2017). Currently, no measure of maladaptive behaviour has been evaluated specifically for its psychometric properties with children with Down syndrome (Esbensen *et al.* 2017).

This study used multimethod assessments of sleep and multi-informant assessments of daytime behaviour to clarify the relationship between common sleep problems and daytime behaviours in children with Down syndrome and expand findings beyond parent report. We focus on sleep problems of shorter sleep duration and poor sleep quality as actigraph measures of these two constructs demonstrate convergent validity with gold standard polysomnography measures of total sleep time and sleep efficiency in children with Down syndrome (Hoffman and Esbensen 2017). We hypothesised that sleep would be related to daytime functioning, specifically that parental report and actigraphy-based measures indicating shorter sleep duration and poorer sleep quality would be associated with parent and teacher reports of higher levels of problematic behaviour. Based on findings with children with intellectual and developmental disabilities and children with Down syndrome, we hypothesised that sleep would be related to parent and teacher reports of inattention, anxiety and challenging behaviour, specifically conduct problems (Harvey and Kennedy 2002; Rzepecka *et al.* 2011). We also examined the relationship between sleep and hyperactivity given the high prevalence of hyperactivity in children with Down syndrome.

Method

Participants

All study activities were approved and overseen by the Institutional Review Board at the medical centre. Community recruitment targeted school-age children with trisomy 21 Down syndrome. Families were recruited through distribution of fliers at the medical centre, a Down syndrome clinic, and newsletters distributed by the local Down syndrome association. Study participants included 30 children with Down syndrome and their parents participating in a community-based pilot study examining sleep and associated daytime behaviour and cognition. One additional family consented but did not complete the study battery. Genetic diagnoses of trisomy 21 were confirmed via medical record. Children with Down syndrome ranged in age from 6 to 17 years (M = 11.68 years, SD = 2.73) and were predominantly male (60%) and Caucasian (93%; 7% African-American). Full-scale IQ on the Kaufman Brief Intelligence Test-2 ranged from 40 to 65 (M = 44.57, SD = 6.46) (Kaufman 2004). The Broad Index score of the Scales of Independent Behaviour-Revised ranged from 15 to 93 (M = 51.26, SD = 21.62) (Bruininks *et al.* 1996). Parent respondents were predominantly mothers (97%). The following medical conditions were reported: congenital heart defect 50%, overweight/obesity 48.2%, recurrent otitis media 36.7%, OSA 33.3%, ADHD 23.3%, GI concerns 20% and anxiety 6.7%.

Measures

Sleep

Sleep duration and quality of sleep were assessed in a multimodal fashion. To obtain objective sleep data, children wore a Micro-Mini Motionlogger Actigraph (Ambulatory Monitoring, Inc.), which is a battery-operated device that closely resembles a watch and measures movement. The actigraph was placed on the nondominant wrist of the participant 30 min before bedtime and removed from the wrist 30 min after rising in the morning. Parents completed a companion sleep diary that was used to corroborate actigraphy estimates. Movement data were processed using a validated sleep scoring algorithm, which differentiates between sleep and wake states (*Micro-Mini Motionlogger Instruction Manual* 2000; Sadeh *et al.* 1994). Specific actigraph measures of sleep duration and sleep quality used for the current analyses included (1) sleep duration: sleep period, the time from when the child fell asleep to when the child woke up, ignoring waking times within that period; and (2) sleep quality: sleep efficiency, defined as the percent of the sleep period that the child spent in

sleep, which offers an objective index of sleep quality. Sleep period and sleep efficiency were determined for each night children wore the actigraph, then averaged across the week to obtain more stable indexes for current analyses. Children demonstrated high compliance during the seven consecutive day actigraph period; 89% of children wore the actigraph for six or seven nights. One child wore the watch for only three nights, and two children wore the watch for four nights. The actigraphs were worn on average for 6.4 nights (SD = 1.1). The mean percentage of nights actigraphs were worn was 91.8%.

While objective, actigraph results are limited to inferences that can be drawn from movement patterns during the recording period. Questionnaires, though collecting subjective impressions, can provide input from a longer term parent observation perspective (Beebe 2012). The CSHQ is a 33-item sleep screening instrument for children and assesses major childhood medical and behavioural sleep disorders during a typical week (Owens *et al.* 2000). Items are rated on a 3-point Likert-type scale from (1) rarely (zero to time/week) to (3) usually (five to seven times/week). Although designed for use in paediatric populations under 10 years of age without intellectual disabilities, the CSHQ demonstrates strong psychometric properties and convergence in identifying behavioural sleep problems in school-age children with Down syndrome ages 6–17 years (Esbensen and Hoffman 2017) and has demonstrated validity in other paediatric populations characterised by intellectual and developmental disabilities (Goldman *et al.* 2012; Richdale and Baker 2014; Veatch *et al.* 2016). Two CSHQ subscales assessing sleep duration and quality were used in the current analyses. The CSHQ includes a three-item sleep duration subscale that assesses parent perception of child's sleep efficiency and consistency. The CSHQ has several other subscales that could relate to sleep quality. After inspection of the most common sleep problems reported in the sample, we elected to use the parasomnias subscale as the parent-reported measure of sleep quality. Although this seven-item subscale incorporates actual parasomnias (e.g. sleep-talking), it also includes an item related to restlessness/movements during sleep that was most often endorsed by parents (83%). Half of parents reported restlessness to occur sometimes, and a third reported restlessness to occur five to seven nights of the week.

Daytime behaviour

Two sets of measures were also used to assess daytime behaviour. We administered the parent and teacher report versions of the Nisonger Child Behaviour Rating Form (NCBRF-P and NCBRF-T) (Aman *et al.* 1996), which was designed specifically for use in populations with intellectual disabilities. Items are rated on a 4-point Likert-type scale from behaviour did not occur or was not a problem (scored 0) to behaviour occurred a lot or was a severe problem (scored 3). This scale demonstrates strong psychometric properties and, unlike some other broad-band behavioural measures, does not directly ask about sleep. The NCBRF demonstrates high inter-rater reliability between parent and teacher forms on all scales and high internal consistency for multiple subscales (Aman *et al.* 1996). The conduct problems and insecure/anxious subscales from the NCBRF were selected based on prior research with children with intellectual disabilities showing relationships between poor sleep and these behavioural domains of challenging behaviour and anxiety (Didden *et al.* 2002; Rzepecka *et al.* 2011).

In order to assess inattention and hyperactivity, which are distinct constructs that could be impacted differentially by sleep difficulties (Beebe 2012), we used the Vanderbilt ADHD Rating Scales – Parent and Teacher Forms (VADPRS and VADTRS). The VADPRS and VADTRS are DSM-IV-based scales that provide clinical information regarding the frequency and severity of symptoms related to ADHD across the home and school domains (Wolraich *et al.* 2003). Internal consistency and reliability are excellent (Wolraich *et al.* 2003) across the nine-item inattention and hyperactivity/impulsivity subscales. Items are rated on a 4-point scale ranging from 0 (never) to 3 (very often) but, as is typical for this measure, a symptom count was tallied for how many items out of nine items were scored 2 or 3 on inattention, and similarly for the nine-item hyperactivity/impulsivity subscale.

Procedure

Children wore an actigraph for seven consecutive nights at home immediately prior to an office visit, during which parents provided information on the child's demographics, daily living skills and completed sleep and behavioural rating forms.

During this office visit, children participated in a neuropsychological battery that included brief IQ testing. During that same week, teachers also completed behavioural rating forms that were distributed and collected by parents. Teacher reports were collected from 25 teachers. Four teacher reports were not obtained as the child was on school break. One teacher report was not returned.

Data analysis

Descriptive statistics and correlational analyses were completed for sleep and daytime behaviour measures. After confirmation that the assumptions of regression (e.g. normality of residuals, multicollinearity of predictors) were not violated, hierarchical linear regressions were used to test whether sleep duration and quality predicted parent and teacher reports of daytime behaviour. Separate regressions were run for each parent-rated and teacher-rated subscale (i.e. NCBRF-conduct, NCBRF-insecure, VADPRS and VADTRS attention and hyperactivity/impulsivity subscales). Age of the child and gender were entered as covariates as sleep concerns vary with age in individuals with Down syndrome and as behaviour concerns vary with gender and age in individuals with intellectual disability (Schroeder *et al.* 1997; Ashworth *et al.* 2013).

Results

Descriptive data for sleep and behaviour measures are presented in Table 1. Descriptive data for the NCBRF are presented as item means, reflecting the total score divided by the number of items on the subscale to support comparison across subscales containing different number of items. Inter-correlations between measures of sleep and behaviours are presented in Table 2. Despite a moderate bivariate correlation between the two CSHQ subscales, multivariate collinearity was found not to be a concern in subsequent regression analyses.

Sleep predicting parent reports of behaviour

Table 3 summarises results of regressions in which sleep measures were entered as predictors of parent-reported daytime behaviours. Neither age nor gender was significantly related to any of the analysed parent report daytime behaviour measures. In contrast, roughly half of the variance on each parent report behaviour measure was statistically predicted by the collective sleep variables (R^2 change = 0.42–0.53, $P < 0.05$). The CSHQ parasomnia subscale was related to the parent-reported conduct problems ($\beta = 0.78$, $P < 0.01$), insecure/anxious behaviours ($\beta = 0.73$, $P < 0.01$), and hyperactivity/impulsivity ($\beta = 0.62$, $P < 0.01$) and had a modest relationship to inattention that did not reach statistical significance. Overall, poorer parent-reported parasomnias were associated with more daytime behavioural concerns. Parent-reported sleep duration was related to inattention ($\beta = 0.40$, $P < 0.05$), but not to other parent reports of daytime behaviour problems. Further, a shorter sleep period, as measured by actigraphy, was related to higher parent ratings of

Table 1 Item mean scores for measures of sleep and behaviour

	Mean (SD)	Range
Sleep measures		
Parent report[†]		
CSHQ sleep duration	4.27 (1.80)	3–9
CSHQ parasomnias	9.38 (2.03)	7–14
Actigraphy[‡]		
Sleep period (minutes)	553.47 (39.37)	466.43–618.64
Sleep efficiency (percent)	87.41 (6.04)	72.49–96.41
Parent behaviour measures		
NCBRF (subscale mean 0–3)[§]		
Conduct problem	0.54 (0.50)	0–2.31
Insecure/anxious	0.18 (0.20)	0–0.81
Vanderbilt (no. of items endorsed 2 or 3)		
Inattention	2.97 (2.86)	0–9
Hyperactivity/impulsivity	1.34 (2.07)	0–7
Teacher behaviour measures		
NCBRF (subscale mean 0–3)[§]		
Conduct problem	0.49 (0.55)	0–2.08
Insecure/anxious	0.24 (0.26)	0–1.13
Vanderbilt (no. of items endorsed 2 or 3)		
Inattention	3.88 (3.11)	0–9
Hyperactivity/impulsivity	1.48 (2.02)	0–7

CSHQ, Children's Sleep Habits Questionnaire; NCBRF, Nisonger Child Behaviour Rating Form.
[†]Possible range of scores on sleep duration is 3–9. Possible range of scores on parasomnias is 7–21.
[‡]Actigraphy measures are averaged over the week the actigraph is worn.
[§]NCBRF scores reflect the total score divided by the number of items to achieve an item mean.

Table 2 Correlations between measures of sleep and behaviour

		1	2	3	4	5	6	7	8	9	10	11
1	CSHQ sleep duration	—										
2	CSHQ parasomnias	0.49**	—									
3	Sleep period	−0.20	0.07	—								
4	Sleep efficiency	0.03	−0.23	0.26	—							
5	NCBRF-P conduct	0.23	0.65**	0.10	0.03	—						
6	NCBRF-P insecure/anxious	0.40*	0.64**	0.06	0.15	0.82**	—					
7	NCBRF-T conduct	0.06	0.62**	0.03	−0.10	0.81**	0.55**	—				
8	NCBRF-T insecure/anxious	−0.03	0.33	0.16	−0.02	0.47*	0.26	0.74**	—			
9	VADPRS inattentive	0.56**	0.49**	−0.35	0.02	0.45*	0.63**	0.42*	0.03	—		
10	VADPRS hyperactive/impulsivity	0.46*	0.55**	−0.39*	0.00	0.73**	0.55**	0.68**	0.38	0.53**	—	
11	VADTRS inattentive	0.09	0.29	−0.14	0.12	0.47*	0.36	0.59**	0.30	0.45*	0.44*	—
12	VADTRS hyperactive/impulsivity	−0.03	0.62**	−0.16	−0.22	0.79**	0.49*	0.82**	0.64**	0.33	0.72**	0.54**

CSHQ, Children's Sleep Habits Questionnaire; NCBRF-P, Nisonger Child Behaviour Rating Form – Parent; NCBRF-T, Nisonger Child Behaviour Rating Form – Teacher; VADPRS, Vanderbilt ADHD Rating Scales – Parent; VADTRS, Vanderbilt ADHD Rating Scales – Teacher.
*$P < 0.05$
**$P < 0.01$.

Table 3 Predicting parent reports of behaviour problems from sleep

	NCBRF-P conduct		NCBRF-P insecure/anxious		VADPRS inattention		VADPRS hyperactive/impulsivity	
	R^2 change	β	R^2 change	β	R^2 change	β	R^2 change	β
Step 1	0.06		0.02		0.07		0.06	
Age		−0.23		−0.13		−0.24		−0.23
Gender		−0.06		0.01		0.08		−0.09
Step 2	0.42*		0.51**		0.53**		0.50**	
Age		−0.01		0.13		−0.10		−0.06
Gender		0.00		0.12		0.31		0.01
CSHQ duration		−0.14		0.09		0.40*		0.04
CSHQ parasomnia		0.78**		0.73**		0.38		0.62**
Sleep period		−0.05		−0.04		−0.36*		−0.50**
Sleep efficiency		0.23		0.31		0.11		0.28

CSHQ, Children's Sleep Habits Questionnaire; NCBRF-P, Nisonger Child Behaviour Rating Form – Parent; VADPRS, Vanderbilt ADHD Rating Scales – Parent.
*$P < 0.05$
**$P < 0.01$.

both inattention ($\beta = -0.36$, $P < 0.05$) and hyperactivity/impulsivity ($\beta = -0.50$, $P < 0.01$), but not conduct problems or insecure/anxious behaviours. Actigraphy-measured sleep efficiency did not significantly predict any of the four parent-reported daytime behaviour scales.

Sleep predicting teacher reports of behaviour

Table 4 summarises results of regressions in which sleep measures were entered as predictors of teacher-reported daytime behaviours. Neither age nor gender was significant predictors of teacher-reported

Table 4 Predicting teacher reports of behaviour problems from sleep

	NCBRF-T conduct		NCBRF-T insecure/ anxious		VADTRS inattention		VADTRS hyperactive/ impulsivity	
	R^2 change	β	R^2 change	β	R^2 change	β	R^2 change	β
Step 1	0.12		0.07		0.17		0.05	
Age		−0.32		0.16		−0.39		−0.26
Gender		−0.15		−0.21		0.15		0.01
Step 2	0.32		0.21		0.08		0.47*	
Age		−0.14		0.34		−0.31		−0.06
Gender		−0.17		−0.27		0.12		−0.05
CSHQ duration		−0.21		−0.16		−0.13		−0.32
CSHQ parasomnia		0.66**		0.50		0.30		0.75**
Sleep period		−0.13		0.02		−0.24		−0.35
Sleep efficiency		−0.11		0.25		0.12		0.07

CSHQ, Children's Sleep Habits Questionnaire; NCBRF-T, Nisonger Child Behaviour Rating Form – Teacher; VADTRS, Vanderbilt ADHD Rating Scales – Teacher.
*$P < 0.05$
**$P < 0.01$.

outcomes. Subsequent entry of the sleep variables again resulted in marginal or substantial prediction of two out of four teacher-reported outcomes. Here, parent report of sleep duration was not predictive of teacher report of daytime behavioural concerns, but more parent-reported problems on the CSHQ parasomnia subscale significantly predicted teacher-reported daytime conduct problems ($\beta = 0.66$, $P < 0.01$) and hyperactivity/impulsivity ($\beta = 0.75$, $P < 0.01$). No sleep index significantly predicted teacher-reported inattention. Actigraphy-based indexes generally were not significantly associated with teacher-reported behavioural concerns.

Discussion

The current study examined the relationship between parent and actigraphy reports of sleep duration and quality with parent and teacher reports of daytime behaviour problems among school-age children with Down syndrome. Parent report of poor sleep quality (as measured by the CSHQ parasomnia subscale), but not actigraph-measured activity during sleep, was predictive of increased conduct problems, insecure and anxious behaviours and daytime hyperactivity/impulsivity. This pattern was similar for both parent-reported and teacher-reported behaviour concerns, indicating that restless sleep as reported by parents has relevance for daytime functioning both at home and at school as observed by independent informants (parent and teacher). These findings parallel findings in children with ASDs, where sleep problems (sleep efficiency, restlessness) are related to oppositional behaviour, anxiety and hyperactivity (Limoges *et al.* 2005; Goldman *et al.* 2009; Mayes and Calhoun 2009). Both parent report and actigraph-measured shorter sleep period was predictive of daytime inattention as reported by parents, and actigraphy-measured sleep period predictive of hyperactivity/impulsivity as reported by both parents. These findings again parallel those in youth with ASD where objective measures of sleep problems (sleep latency) are related to an omnibus indicator of maladaptive behaviour problems (Bruni *et al.* 2007; Goldman *et al.* 2009). The present findings support the previous literature that sleep problems are related to daytime behaviour concerns in children with Down syndrome, in children with intellectual and developmental disabilities (Richdale *et al.* 2000; Didden *et al.* 2002) and children with ASDs (Goldman *et al.* 2011; Sikora *et al.* 2012).

These correlational findings cannot definitively establish causation – i.e. that sleep problems cause daytime behaviour problems or that daytime behaviour problems contribute to poor sleep. Further research is needed to determine causality and inform

interventions. However, based on the literature from typically developing children, others suggest that insufficient sleep duration or quality may serve as a setting event for noncompliant behaviours displayed at home and at school (Didden *et al.* 2002; Stores *et al.* 1998; Wiggs and Stores 1996). Thus, interventions to improve daytime behaviour may need to also target or address current sleeping patterns. Communication between home and school regarding current sleeping patterns may be a helpful tool to understanding the role of sleep in behaviours observed at school and the implementation of appropriate behavioural strategies. Empirically supported interventions for behaviours associated with sleep and sleep hygiene may be beneficial to improving sleep duration and quality. Currently, the first randomised clinical trial is underway to evaluate behavioural sleep strategies for improving sleep in children and adolescents with Down syndrome (NCT02996175).

Current findings also highlight that the relationship between sleep problems and behaviour problems can vary depending on how sleep problems are assessed, using parent reports or actigraphy. Indeed, despite our intent to perform multimodal assessments of the same constructs, it is noteworthy that parent-reported sleep duration correlated poorly with actigraphy-measured sleep period, while parent-reported sleep disruption (as measured by questions about nocturnal restlessness and parasomnias) correlated poorly with actigraphy-measured sleep efficiency or activity during sleep. Future research is needed to better understand this lack of correlation. Potential causes could include errors in parental estimates (e.g. due to selective awareness of events that disrupt parents' sleep), movement-related artefacts on actigraphy in children with Down syndrome (a possibility that has not yet been examined) or subtle but important differences in the constructs being measured. On the latter point, whereas actigraphy provides an objective estimate of sleep duration, the CSHQ sleep duration subscale also asks about consistency of sleep length and perception of sleep relative to appropriate sleep length. This introduces an element of parental subjectivity and judgement that may reduce associations with daytime behaviours. Conversely, whereas actigraphy-measured sleep efficiency or activity during sleep is affected by both brief and lengthy waking bouts, it may be that parental report on the CSHQ assesses aspects of sleep disruption that are more selectively related to daytime behaviours. It may not be that one measurement source is 'better' than the other, but that each has unique information to contribute. These findings underscore the importance of measuring behavioural sleep problems with multiple methods that are hypothesis-driven.

The sample in this pilot study was small and presented with a low rate of behavioural concerns, and thus provided limited power for conducting additional statistical analyses. For example, with this smaller sample size, it would be difficult to evaluate the impact of medical comorbidities on the relationship between sleep and daytime behaviour problems. In addition, although the CSHQ demonstrates convergent validity with polysomnography, the gold standard for assessing sleep problems remains assessing sleep with polysomnography (Hoffman and Esbensen 2017). Despite these limitations, our findings replicate and extend the initial findings in the literature of a relationship between sleep problems and daytime behaviour in children with Down syndrome. Further work is needed to explore the bidirectional relationship between behaviours and how they may impact sleep. Our findings demonstrate a pattern of poor sleep contributing to a pattern of poor behaviour. Further work is needed to explore the immediate relationship between sleep and daytime behaviour on the following day. Future studies would benefit from examining the impact of medical comorbidities (obesity, OSA, ADHD and ASDs) on the relationship between sleep and daytime behaviour problems.

Children with Down syndrome are at increased risk for sleep problems (Esbensen and Schwichtenberg 2016). Our study corroborates that sleep problems are a concern and also that these sleep problems are associated with daytime problem behaviours, particularly conduct behaviours, anxious behaviours, inattention and hyperactivity/impulsivity. Thus, while screening for OSA is well-recognised in the Down syndrome community (Bull and Genetics 2011), there also is a need for appropriate screening for other sleep problems in children with Down syndrome, as well as a need for behavioural interventions to understand the role of sleep in contributing to these behaviours. Future research is needed to understand how sleep problems may hinder behavioural interventions and

how sleep interventions may improve daytime behaviours.

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