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Brief report

Parental report of sleep problems in Down syndrome

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Abstract

Background Children with Down syndrome (DS) suffer from sleep problems, including sleep maintenance problems, as well as snoring, and other symptoms of disordered breathing. To examine sleep in DS, we gave parents a questionnaire assessing their child's sleep.
Materials and methods The parents of 35 children with DS (mean age = 12.65 years, range = 7–18 years) completed the 33-item Children's Sleep Habits Questionnaire.
Results Eighty-five per cent of our sample had sleep disturbance scores in the clinical range (mean = 48.63, SD = 7.15, range = 34–64). Our sample also had significantly elevated scores on the Bedtime Resistance, Sleep Anxiety, Night Wakings, Parasomnias, Sleep Disordered Breathing and Daytime Sleepiness subscales.
Conclusions Children with DS are at risk for developing symptoms of sleep disordered breathing, and may have additional sleep problems that are unrelated to sleep disordered breathing.

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Keywords Down syndrome, intellectual disability, parents, sleep studies

Introduction

Sleep problems are not uncommon among elementary school-aged children (Owens *et al.* 2000). However, they are far more prevalent in children with intellectual disabilities. Specifically, sleep disturbance is frequently reported by parents of their children with Down syndrome (DS). Problems with bedtime settling, sleep onset, sleep maintenance and early morning waking as well as snoring and other symptoms of sleep disordered breathing have been observed (Stores *et al.* 1998; Levanon *et al.* 1999; Cotton & Richdale 2006). Laboratory polysomnographic studies have reported the presence of obstructive sleep apnoea (OSA) in this population to be between 30% and 79% (Dyken *et al.* 2003; Shott *et al.* 2006).

Sleep disturbance has a serious impact on children with DS as well as their families, caregivers, teachers and peers. Problems with sleep have been shown to negatively impact cognitive function (Beebe *et al.* 2004; Blunden *et al.* 2005), academic performance (Gozal 1998) and behaviour (Chervin *et al.* 2002). In addition, sleep disturbance has been

related to parental sleep deprivation, relationship difficulties and maternal depression (Pain 1998; Lam *et al.* 2003).

Previous studies have administered sleep questionnaires to parents of children with DS, and found that parents of preschool- and school-aged children consistently reported clinically significant sleep problems across sleep domains. Prior to the validation of the Children's Sleep Habits Questionnaire (CSHQ), Stores *et al.* (1998) administered a 49-item sleep questionnaire (adapted from Simonds & Parraga 1982) to 91 parents of children with DS (mean age = 10.5 years) that assessed a wide range of potential sleep problems. They derived three sleep disturbance factors from a principal components analysis: (1) sleep onset problems; (2) sleep maintenance problems; and (3) sleep disordered breathing.

Two studies (Carter *et al.* 2009; MacCrosain & Byrne 2009) administered the CSHQ to school-aged children with DS (mean age = 8.6 years and 7.6 years, respectively) and found that nearly all (96.8–100%) of their participants had total sleep disturbance scores in the clinical range. Carter *et al.* (2009) found that all eight subscales were significantly abnormal in children with DS aged 4–12 ($n = 24$) compared to data published for typically developing children aged 4–10. Older children (≥13 years, $n = 16$) with DS had significantly elevated scores on the bedtime resistance, sleep duration, parasomnias, sleep disordered breathing and daytime sleepiness subscales. MacCrosain & Byrne (2009) found that children with DS scored higher on all eight subscales compared to published data for typically developing children aged 4–10.

In the present study, we administered the CSHQ to a cohort of children with DS in the USA, which is considerably older on average than previous cohorts. Using a cross-sectional sample, we hoped to (1) compare the data obtained for our group of older children with DS with the data of younger children with DS as well as with the data of younger typically developing children, and (2) determine any correlation between sleep disturbances and age within our group of children with DS. Based on the predisposition that children with DS have for developing OSA and the prevalence estimates in the literature, we anticipated that OSA-related symptoms would persist throughout childhood. Furthermore, we expected fewer reports of problems with bedtime resistance, sleep onset and maintenance, sleep anxiety and parasomnias (e.g. night terrors, nightmares, etc.) in our population than in previous studies with younger children with DS, as these problems tend to decline with age in typically developing children.

Materials and methods

Participants

The parents of 35 children medically diagnosed with DS (mean age = 12.65 years, SD = 3.55, range = 7–18 years; 16 girls) completed the CSHQ at one time-point as part of a larger research project investigating the relationship between sleep and cognition in DS. Subjects were recruited through local support group networks and with newspaper advertisement. Recruitment was not directed towards the study of sleep disorders, and this may have reduced bias towards sleep disturbance among those who volunteered to participate. Our sample was ethnically diverse and from a middle-class background (see Table 1).

Procedure

All study procedures were approved by the human subjects committee at the University of Arizona. Parents completed the CSHQ either at their home or in our laboratory alongside other questionnaire measures regarding their child.

Table 1 Demographic characteristics of children with Down syndrome ($n = 35$)

Mean age in years (SD)	12.65 (3.55)
Age range	7.50–18.83
Gender girls (%)	16 (45%)
Karyotype	28 Trisomy 21, 1 Robertsonian Translocation, 1 Mosaicism, 5 unknown
Ethnicity	16 European American, 16 Hispanic, 1 Asian American, 2 unspecified
Mean total income	$62 200

Measures

The Children's Sleep Habits Questionnaire

The CSHQ is a 33-item screening instrument that yields both a total score (total possible = 103, range = 33–103) and eight subscale scores, reflecting key sleep domains that encompass the major medical and behavioural sleep disorders in school-aged children. Subscales include Bedtime Resistance, Sleep Onset Delay, Sleep Duration, Sleep Anxiety, Night Waking, Parasomnias, Sleep Disordered Breathing and Daytime Sleepiness. The CSHQ has been used previously to assess sleep problems in DS populations (Shott *et al.* 2006; Carter *et al.* 2009; MacCrosain & Byrne 2009) and has been validated in school-aged children (i.e. 4–10 years; Owens *et al.* 2000).

Power analyses

We conducted power analyses for our planned significance tests. Findings from previous studies (Carter *et al.* 2009; MacCrosain & Byrne 2009) allowed us to anticipate a large effect for our comparison of means, with the effect sizes in these studies ranging from $d = 0.65$ to $d = 1.8$. Based on Cohen's (1992) tables, there is sufficient power (0.80) to detect a large effect with a total sample size of 35 and $\alpha = 0.05$ for one sample *t*-tests and simple regression.

Results

We found that 85.7% of our sample had total sleep disturbance scores in the clinical range, that is, greater than 41 (mean = 48.63, SD = 7.15, range = 34–64). Twenty per cent of our sample regularly needed a parent in the room in order to fall asleep, and 28.5% fell asleep in a parent or sibling's bed at least two nights per week. Thirty-four per cent of our sample had difficulty falling asleep within 20 min of going to bed at least two nights per week. Once asleep, 43% were described as occasionally restless, while 37% were described as usually restless during sleep. Commonly described parasomnias included bruxism, or grinding of the teeth, which occurred at least two nights per week in 34% of children and sleep talking, which occurred in 29%. Sixty per cent of our sample endorsed at least one symptom of sleep disordered breathing, including loud snoring, cessation of breathing, and snorting and gasping during the night. Importantly, each daytime sleepiness item was endorsed by at least 25% of our sample. Over half of our sample routinely fell asleep while watching television (57%) or while riding in the car (60%).

Compared to published data for typically developing children in a community sample aged 4–10 years, our sample also had significantly elevated scores on the Bedtime Resistance [$t(34) = 2.36$, $P = 0.02$], Sleep Anxiety [$t(34) = 2.76$, $P < 0.01$], Night Wakings [$t(34) = 3.33$, $P < 0.01$], Parasomnias [$t(34) = 4.51$, $P < 0.001$], Sleep Disordered Breathing [$t(34) = 4.05$, $p < 0.001$] and Daytime Sleepiness [$t(34) = 4.617$, $P < 0.001$] subscales (see Fig. 1). We found a trend towards elevated scores on Sleep Onset Delay subscale [$t(34) = 1.75$, $P = 0.08$] and no significant difference on the Sleep Duration subscale [$t(34) = 0.905$, $P = 0.37$].

We also examined the relationship between chronological age and subscale or sleep disturbance scores in this sample using linear regression. Age was significantly related to the Sleep Anxiety subscale, with lower scores in older versus younger children ($F_{1,34} = 5.79$, $P = 0.02$, $\beta = -0.39$). The

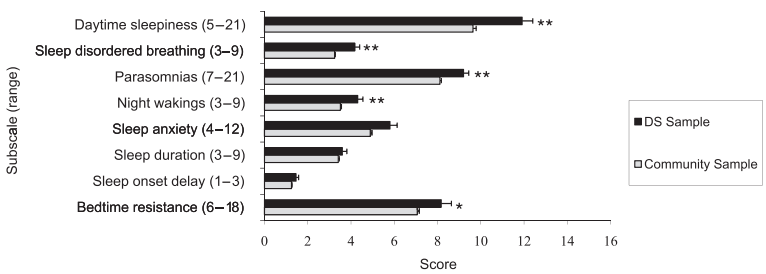

Figure 1 Children's Sleep Habits Questionnaire subscales: comparison of children with Down syndrome (DS) and a community sample of typically developing children. Statistical difference at the level of $*P < 0.05$, at the level of $**P < 0.01$. Error bars represent standard errors of the means.

younger children's scores were significantly elevated compared to the published data (Owens *et al.* 2000), while the older children's scores were not. There were trends for significant differences in other clinical sleep complaints across age, including decreases in Bedtime Resistance ($F_{1,34} = 3.29$, $P = 0.08$, $\beta = -0.30$) and Parasomnias ($F_{1,34} = 3.87$, $P = 0.06$, $\beta = -0.32$), and increases in Sleep Onset Delay ($F_{1,33} = 2.97$, $P = 0.09$, $\beta = 0.29$). There were no differences between younger and older children on any other subscale or on the overall sleep disturbance score.

Discussion

This study found that sleep problems are quite common in a community sample of school-aged children with DS aged 7–18 years. These findings suggest that sleep problems do persist in this population. Given that sleep is important to memory, learning and behaviour in typically developing children, interventions for sleep problems are clearly warranted across a wide range of ages in DS.

When compared to Owens *et al.*'s (2000) community sample (Fig. 1), our sample scored higher on six of the eight subscales. These findings are similar to those of MacCrosain & Byrne (2009) and Carter *et al.* (2009). While MacCrosain and Byrne reported higher scores in children with DS across all eight subscales, Carter *et al.* (2009) found that parents from an older subset of their sample (mean age = 16.3, $n = 16$) reported sleep problems on the Bedtime Resistance, Parasomnias, Sleep Disordered Breathing and Daytime Sleepiness subscales. In our sample, there was a trend for parents of children with DS to report longer sleep latencies (Sleep Onset Delay) than parents from the Owens *et al.* (2000) community sample. While parents of children with DS had more concerns about their child's sleep duration than parents from the community sample, this difference was not significant. This finding may reflect a developmental difference, such that adolescents with DS have a total sleep time that parents perceive to be appropriate for their age whereas younger children with DS are perceived to sleep 'too much' or 'too little.'

Elevations on the Bedtime Resistance, Sleep Anxiety and Night Wakings subscales are consistent with previous literature describing settling problems and difficulties with sleep onset and sleep maintenance in this population (Stores *et al.* 1998; Cotton & Richdale 2006). Children experiencing these difficulties may benefit from behavioural sleep medicine interventions, such as sleep education, positive daily and bedtime routines and sleep hygiene, and graduated extinction techniques (Reid *et al.* 2009). The Bedtime Pass Program, which is an example of an extinction-based procedure for treating bedtime resistance (Moore *et al.* 2007), has been shown to be effective in typically developing children in reducing the time to fall asleep, as well as the number of cries for parents during the night, and the number of times leaving the bedroom, and these effects were maintained at 3-month follow-up. Although it has not yet been systematically evaluated in children with developmental or intellectual disabilities, it is a non-complex intervention that parents have found to be highly satisfactory.

Elevated scores on the Sleep Disordered Breathing and Excessive Daytime Sleepiness subscales are consistent with the high rates of OSA that have been reported and confirmed with polysomnography in children with DS (Dyken *et al.* 2003; Shott *et al.* 2006). Children with DS who experience symptoms of sleep disordered breathing should have a sleep assessment performed as early as possible (American Academy of Pediatrics 2001). If they meet criteria for an OSA diagnosis, most children with DS have an adenotonsillectomy. Children with DS often experience a reduction in their OSA symptoms but frequently still meet criteria for OSA after surgery (Merrell & Shott 2007). In children with DS whose OSA falls into the mild range, positional treatments and mandibular advancement devices may be useful, although these devices have not been systematically evaluated in this population. In addition, the mandibular advancement device might also assist in reducing bruxism. Shires *et al.* (2010) have shown that body mass index has a statistically significant association with the presence of OSA in children with DS, which suggests that weight loss may also help to reduce the incidence and severity of OSA in this population. In children with DS with moderate to severe OSA, nasal continuous positive airway pressure (nCPAP) has been shown to be an effective treatment. O'Donnell *et al.* (2006) found that 72% of children with DS

($n = 22$) accepted and adhered to nCPAP treatment.

The Parasomnias subscale was also elevated in our sample. This is primarily due to parents endorsing items about restlessness, bruxism and sleeptalking. The restlessness and bruxism may be related to the arousals associated with OSA (Oksenberg & Arons 2002).

When we performed a linear regression across chronological age for each subscale and the total sleep disturbance score, we found that Sleep Anxiety subscale scores declined with increasing age, as has been previously reported in typically developing children. These results suggest that problems with bedtime resistance, parasomnias, night wakings, sleep disordered breathing and daytime sleepiness persist across development, while sleep anxiety problems tend to decline across childhood in DS. Future studies examining change in levels of sleep disturbance over time within a DS cohort would provide robust evidence for this conclusion.

In conclusion, our study suggests that children with DS have sleep problems that include bedtime resistance, sleep onset problems, sleep maintenance problems and sleep disordered breathing and extends previous findings into adolescence. These findings underscore the need for both behavioural sleep medicine interventions as well as treatment for sleep disordered breathing in this population.

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