SLEEP AND COGNITION IN PRESCHOOLERS WITH AND WITHOUT DOWN SYNDROME

Sleep-disordered breathing and cognitive functioning in preschool children with and without Down syndrome

Short title: Sleep and cognition in children with and without Down syndrome

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Abstract

Background: Sleep affects children's cognitive development, preparedness for school and future academic outcomes. People with Down syndrome (DS) are particularly at risk for sleep disordered breathing (SDB). To our knowledge, the association between SDB and cognition in preschoolers with DS is unknown.

Methods: We assessed sleep using cardiorespiratory polygraphy in 22 typically developing (TD) preschoolers, and 22 with DS. Cognition was assessed using the Mullen Scales of Early Learning, and behaviour using the Strengths and Difficulties Questionnaire (SDQ). The MacArthur Communicative Development Inventory (MCDI) measured language level. We predicted that sleep problems would be associated with lower cognitive and behavioural functioning.

Results: In TD children, longer sleep duration was associated with higher scores on MCDI expressive language, and fewer emotional symptoms such as fear and unhappiness on the SDQ; whilst SDB was associated with increased conduct problems and less prosocial behaviour on the SDQ. Conversely, for children with DS, SDB was associated with increased language understanding and use of actions and gestures on the MCDI.

Conclusions: The findings in the TD group support our hypotheses. We recommend that sleep problems are screened for and treated as even mild SDB may prompt poorer cognition and behaviour. For children with DS, we expect that multiple factors in this complex syndrome mask or mediate the association between sleep and cognitive development, and tighter controls are necessary to uncover effects of sleep. We propose longitudinal studies as a necessary tool to assess the precise impact of sleep on cognitive development in accounting for individual differences in DS.

Key words: sleep, sleep disorders, cognition, behaviour problems, Down syndrome, children

Abbreviations: Strengths and Difficulties Questionnaire (SDQ); MacArthur Communicative Development Inventory (MCDI); Down syndrome (DS); typically developing (TD), obstructive sleep apnoea syndrome (OSAS); peripheral oxyhaemoglobin (SpO_2).

SLEEP AND COGNITION IN PRESCHOOLERS WITH AND WITHOUT DOWN SYNDROME

Sleep is vitally important for children's healthy development. Around 20-30% of infants and children experience some kind of sleep problem (Mindell, Kuhn, Lewin, Meltzer, & Sadeh, 2006) which may have significant, wide-reaching detrimental effects if left unmanaged.

The current study focuses on sleep-disordered breathing (SDB), which covers a range of symptoms from primary snoring (snoring in the absence of associated respiratory events) to obstructive sleep apnoea syndrome (OSAS), where the upper airway becomes blocked, causing difficulty breathing during sleep. OSAS often leads to fragmented sleep as apnoeas (pauses or obstructions in breathing), hypopnoeas (abnormally shallow breathing) and associated decreases in oxygen (hypoxia) and increases in carbon dioxide (hypercarbia) can lead to night wakings (American Academy of Sleep Medicine, 2001; Thorpy, 2012). OSAS is thought to affect around 2% of children (Brunetti et al., 2001) and is associated with a range of physiological (Tauman & Gozal, 2011) and cognitive deficits. For example, preschoolers who are reported to snore have more difficulties with planning, inhibition and working memory compared with non-snorers (Karpinski, Scullin, & Montgomery-Downs, 2008). Bourke and colleagues (2011) assessed 137 children with polysomnography and a range of cognitive tests. Healthy children had higher verbal and full scale IQ on the Wechsler Abbreviated Scale of Intelligence than children in all SDB groups (primary snoring, mild, and moderate/severe OSAS), but performance IQ did not differ by group. Children with SDB were more likely to be impaired on executive and academic tasks (reading and arithmetic), but this was not related to the severity of SDB. Similarly, Blunden, Lushington, Lorenzen, Martin, and Kennedy (2005) found children who reportedly snored or had behavioural sleep problems had lower full scale and verbal IQ than healthy children, whilst performance IQ was no different.

In a review, Ednick et al. (2009) discussed objective and parent-report studies of infants in their first year of life and consistently found that sleep duration, quality and stability were associated with improved mental and psychomotor development. However, it is still unclear whether sleep characteristics at this early age cause developmental outcomes, or whether brain maturation

underpins both facets. Nevertheless, the consequence is that children who experience sleep problems have poorer cognitive abilities at school entry, including attentional and executive function deficits, poorer motor skills and reduced language abilities compared to children who sleep well (Karpinski et al., 2008).

Down Syndrome

Down syndrome (DS) is the most common sporadic genetic disorder, occurring in 1 in 700-1000 live births (Parker et al., 2010; Roizen & Patterson, 2003). It is caused by an additional copy of chromosome 21 (trisomy 21) and results in characteristic physical symptoms and intellectual impairment with great inter-individual variability (Glasson et al., 2002).

Sleep problems are frequently reported in individuals with DS, including difficulties initiating and maintaining sleep, early morning waking and daytime sleepiness (Richdale, Francis, Gavidia-Payne, & Cotton, 2000; Tietze et al., 2012). In addition, several contributory factors predispose them to OSAS, including craniofacial and upper airway abnormalities, obesity, tonsil and adenoid encroachment, macroglossia, generalised hypotonia and recurrent upper respiratory tract infections (Churchill, Kieckhefer, Landis, & Ward, 2012). OSAS is thought to affect around 80% of children with DS (Austeng et al., 2014). Sleep problems are often under-recognised and rarely reported by parents, whereas physicians tend to assume sleep problems to be simply a feature of DS (Shott et al., 2006). Few studies have investigated associations between sleep and cognition in DS. In 38 children with DS, Breslin et al. (2014) found that those who experienced OSAS had a 9-point-lower verbal IQ score and poorer cognitive flexibility than children without OSAS. OSAS has also been linked with visuo-perception problems in 12 young adults with DS (Andreou, Galanopoulou, Gourgoulianis, Karapetsas, & Molyvdas, 2002). Parentally-reported OSAS symptoms have also been linked with poorer verbal fluency and inhibition in 29 adolescents and young adults with DS (Chen, Spanò, & Edgin, 2013). In 29 toddlers with DS, those with low sleep efficiency as measured by

actigraphy (movement monitoring) had poorer expressive language skills relative to those with high sleep efficiency (Edgin et al., 2015). In general, these studies suggest a particular association between sleep problems and language difficulties for people with DS, which echoes TD data.

To our knowledge, no studies have yet examined OSAS in relation to early cognitive development in DS. This is an important and challenging area since early development is predictive of future developmental outcomes. The aim of the current study was to assess the association between SDB and cognitive and behavioural abilities in 2- to 4-year-olds with DS compared to age-matched TD children. We hypothesised that 1) children with DS would experience increased SDB-related symptoms relative to TD children; 2) children with DS would have lower cognitive and behavioural scores; 3) improved scores would be associated with fewer SDB symptoms, with a particularly strong association between sleep and language abilities for both groups.

Methods

Participants

Twenty-three TD children (14 male) and 22 children with DS (16 male), group matched for age, took part in the study. One TD boy (age 30.46 months) was later excluded as he did not tolerate wearing sleep-monitoring equipment and his parent did not return the questionnaires. TD children were recruited through local nurseries, newsletters and social media, whilst children with DS were recruited through parent groups including the Down's Syndrome Association, social media, and the local special educational needs and disabilities service. Parents responded to an advert to take part in a study on sleep and cognitive development. With the aim of recruiting children across the range of respiratory health, around half of the TD adverts stated that we were looking for 'children who snore'. Parents were provided with full information in writing. Details of the final sample are presented in Table 1. All children were healthy at the time of the study, had no comorbid disorders and were not taking hypnotic medication. Hearing and vision were normal or corrected to normal. All

children were singletons and were at least 35 weeks gestational age at birth, although children with DS were born earlier than TD children. An independent samples *t*-test and Chi-square respectively showed no age or sex differences between groups.

Parental education was measured as a possible confounding variable. Parents reported their highest qualification level and this was standardised as a measure of years in post-16 full-time education based on length of courses in the UK (e.g. GCSE=0, A-level=2, Bachelor's degree=5 total years). There was no significant difference between groups for maternal or paternal education.

Table 1

The study was approved by XXX Research Ethics Committee. All parents gave written informed consent. Assent from the child was gained verbally where possible, and was otherwise assumed based on cooperation. Parents were given a £40 shopping voucher for taking part as reimbursement for time and travel expenses.

Materials

Cognitive and Behavioural Assessment. Children completed the Mullen Scales of Early Learning which is a standardised developmental test assessing five domains: fine motor, gross motor, visual reception, expressive language and receptive language (Mullen, 1995). Performance on the Mullen Scales correlates well with other measures of early development, e.g., the Bayley Scales of Infant Development, and the Peabody Fine Motor Scale; however, we selected the Mullen as it has high internal consistency (ranging from .75 to .83 for each scale) and due to its coverage of both verbal and non-verbal skills, its suitability for the full developmental age range of interest, and its previous successful use for children with DS (Fidler, Hepburn, & Rogers, 2006; Mullen, 1995; Roberts & Richmond, 2015).

Children were seated independently or on their parent's lap at a table opposite the researcher (XXX). A parent was always present and was asked not to intervene with the testing other than to offer reassurance to the child. Children were encouraged to remain at the table but, if this proved too difficult, some tasks were carried out on the floor or at a low table until they returned to their seat. Children were given frequent breaks, as necessary.

Questionnaires. The Strengths and Difficulties Questionnaire for 2-4 years (SDQ) (Goodman, 1997), a brief 25-item screening tool, was used to assess behavioural functioning. Parents report whether each behaviour is certainly, somewhat, or not true of their child. The SDQ yields scores on five subscales (emotional symptoms, conduct problems, hyperactivity, peer problems and pro-social behaviour) as well as an impact score for how much the child's difficulties affect their daily life. Higher scores indicate increased expression of each subscale.

The MacArthur Communicative Development Inventory (MCDI) – Words and Gestures (Fenson et al., 1993) is a standardised checklist for language development in children aged <30 months, measuring parents report of receptive and expressive language and use of gestures. Parents check whether their child understands each word, or understands and can also say each word, and whether their child uses gestures or mimics actions.

Sleep Assessment. OSAS was assessed with domiciliary cardiorespiratory polygraphy for one night using the SOMNOtouch™ (SOMNOmedics, Germany). This body-worn device comprises abdominal and thoracic respiratory inductance plethysmography (RIP); pulse oximetry (Noonin/Masimo) yielding peripheral oxyhaemoglobin (SpO_2) and pulse; nasal pressure flow with snore sensor; body position sensor and actigraphy. It has previously demonstrated a success rate of 93% in children with DS (Hill et al., 2016) and is an acceptable alternative to polysomnography for diagnosis of sleep apnoea (Kaditis et al., 2016). Domino Light software (SOMNOmedics, Germany) was used for scoring according to AASM paediatric scoring criteria. Where the nasal flow signal was lost, RIP sum was used as the recommended alternative measure of airflow (Berry et al., 2012).

SLEEP AND COGNITION IN PRESCHOOLERS WITH AND WITHOUT DOWN SYNDROME

Variables of interest were key indicators of SDB, including number of apnoeas and hypopnoeas, baseline SpO_2 and SpO_2 desaturations. Parents reported their child's usual total sleep duration, including daytime and night time sleep.

Procedure

Children individually attended the research centre at XXX University with a parent. Parents were advised to attend at a time when their child would be most likely to be wide awake, and not coinciding with habitual nap times; thus, sessions were usually in the morning. Children completed the Mullen Scales of Early Learning. Parents were trained in use of the SOMNOtouch™ and RIP bands measured to fit the participant. Parents were given printed written and photographic instructions and a contact telephone number. They were requested to record for one full night if possible and were told that at least four hours of artefact-free recording (i.e., good quality recording during sleep without movement or interference) were necessary for analysis (Urschitz, Brockmann, Schlaud, & Poets, 2010). Questionnaires were explained, and questions were answered. The session usually took around 1:30 hours. Parents completed the domiciliary sleep study, usually on the night of the cognitive tests and always within one week. Questionnaires were completed at home. Parents returned the equipment and questionnaires either in person or by courier within one week. Occasionally parents needed to repeat the study: in three cases equipment was returned with insufficient recording and parents repeated successfully within two weeks of the child's cognitive assessment. Parents were given a report on their child's sleep with the caveat that a research study should not be used for clinical diagnosis.

Analysis

Data were analysed using IBM SPSS v 22 and inspected for outliers visually and using Cooks distance. Where outlying scores changed the significance of findings they were removed, otherwise they were included (Thomas et al., 2009).

Groups (TD and DS) were compared using independent samples t-tests. Levene's test was used to test for homogeneity of variance and where this was violated test statistics are reported for unequal variances. Chi-square was used to assess group differences on categorical variables.

Hierarchical multiple linear regression was used to test relationships between sleep parameters and performance on each Mullen, SDQ, and MCDI scale. We first assessed data for possible confounding factors. Pearson's correlation coefficient showed chronological age to be significantly related to performance on almost all variables. We thus controlled for age when investigating mother's education, father's education, and age at gestation using partial correlations; and sex using ANCOVA. We found sex differences for some areas of performance and control for these when present. Parental education and age at gestation were not significant confounders; thus we did not enter them into the models.

Predictor variables were entered into the model in blocks. Block one always controlled for chronological age. Block two controlled for sex, only if sex had an influence on performance after controlling age. In block three we tested our sleep variables of interest. Since we did not know precisely which variables would have the strongest associations with behaviour, and in order to eliminate unnecessary predictors, we entered block three using a stepwise approach with the following predictors: desaturation index, apnoea/hypopnoea index, apnoea index, minimum SpO_2, baseline SpO_2, and total sleep time as reported by parents.

The Durbin-Watson statistic assessed the assumption of independent errors, and standardised residuals and predicted values were inspected visually for normality. Tables include standardised Beta values (β) alongside Beta (B) since they are more comparable than non-standardised values.

Finally, to assess potential non-linear relationships demonstrated in other studies (Bourke et al., 2011; Breslin et al., 2014), a median split was performed on AHI for the TD (median=0.60) and

the DS (median=1.10) groups. There were no age or sex differences between high and low AHI

groups so we used *t*-tests to compare their performance on each Mullen, SDQ and MCDI scale.

Results

Mullen Scales of Early Learning

As predicted, TD children performed significantly better than children with DS across all

areas of the Mullen Scales (Table 2). Raw scores on the task were converted into mental ages (in

months) as per the manual. We report total score as a raw score since the Gross Motor scale is capped

at 33 months; thus, a ceiling effect in the TD group would lower mean scores if we averaged the

scores of each scale.

Table 2

Strengths and Difficulties Questionnaire

Scores for each subscale were calculated by summing items, allowing a possible score range

from 0 to 10. Two children with DS (male age 38.60 months, and female age 34.89 months) had

incomplete questionnaires, thus have some subscales missing.

As hypothesised, in comparison to the TD group, children with DS had significantly more

peer problems and exhibited less prosocial behaviour and their difficulties had more impact on daily

life (Table 3).

Table 3

MacArthur Communicative Development Inventory

Scores were summed to give a total count for words that a child Understands, Understands

and Says; and Actions and Gestures used by the child. Data were missing for five TD boys and two

TD girls (age range 31.51 to 52.44 months) whose questionnaires were either not returned or were

not complete. In support of our hypothesis, *t*-tests showed that TD children scored significantly

higher than children with DS on all scales (Table 4).

Table 4

Cardiorespiratory Polygraphy

Groups were compared for key cardiorespiratory variables using indices (the number of

events per hour of recording) (Table 5). Data for one TD girl (age 27.93 months) were removed as

recording time was under one hour (0:43) so indices could not be calculated accurately. Two further

TD children (Male, age 39.62 months; female, age 48.23 months) with artefact-free recording

durations of 2:23 and 3:04 hours respectively were included in analyses because their inclusion did

not change the significance of findings. Only nine TD and seven children with DS tolerated wearing

the nasal cannula for the full duration of the study, and three TD and two children with DS partially

tolerated it, so we have not reported snoring due to small sample sizes.

As predicted, children with DS had increased evidence of SDB, shown by a higher

hypopnoea index, apnoea/hypopnoea index (AHI), 3% desaturation index, baseline SpO_2 and

minimum SpO_2. They also had a longer total and daytime sleep duration as reported by parents.

Table 5

Association between sleep and performance on the Mullen Scales of Early Learning

Contrary to our hypothesis, regression models showed that sleep variables were not able to predict performance for either group for Mullen Scales of Gross Motor, Fine Motor, Visual Reception, or Receptive Language skills in either group. In the TD group, lower minimum SpO$_2$ was associated with poorer scores on the Expressive Language scale (ΔR^2=.11, $\Delta F(1,15)$=7.58, p=.02) after controlling age in block one (R^2=.55, $F(1,17)$=21.12, p<.001) and sex in block two (ΔR^2=.11, $\Delta F(1,16)$=5.05, p=.04). Appendix Table 1 shows the results of the regression models for both groups on the Expressive Language Mullen Scale.

Association between sleep and behaviour reported by parents on the SDQ

We found several associations between sleep characteristics and behaviour reported by parents for the TD group but not for the DS group.

As predicted, for TD children greater emotional problems were associated with shorter sleep duration (ΔR^2=.57, $\Delta F(1,16)$=23.68, p<.001) after controlling for age (R^2=.04, $F(1,17)$=0.69, p=.42); increased conduct problems were associated with a higher AHI (ΔR^2=.29, $\Delta F(1,16)$=6.97, p=.02) after controlling for age (R^2=.05, $F(1,17)$=0.95, p=.34); and increased prosocial behaviour was associated with a lower apnoea index (ΔR^2=.25, $\Delta F(1,16)$=6.80, p=.02) after controlling for age (R^2=.16, $F(1,17)$=3.19, p=.09). Also in the TD group, a higher total problem score was associated with increased AHI (ΔR^2=.26, $\Delta F(1,14)$=4.85, p=.045) after controlling for age (R^2<.001, $F(1,15)$=0.001, p=.97). Appendix Tables 2-5 show the results of regression models for both groups where associations with sleep were found. Sleep characteristics were not associated with Hyperactivity, Peer Problems, or Impact for either group.

Association between sleep and language reported by parents on the MCDI

Regression models showed that TD children with longer sleep duration exhibited higher scores on the Understands and Says scale of the MCDI (ΔR^2=.23, $\Delta F(1,9)$=6.27, p=.03) after

controlling for age (R^2=.43, $F(1,10)$=7.70, p=.02). Only 12 TD children were included in this model due to missing data.

Contrary to our hypotheses, for children with DS higher scores on the Understands scale were associated with a lower baseline SpO$_2$ (ΔR^2=.18, $\Delta F(1,19)$=12.67, p=.002) after controlling for age (R^2=.55, $F(1,20)$=24.41, p<.001); and higher scores for Actions and Gestures were associated with a higher desaturation index (ΔR^2=.14, $\Delta F(1,19)$=6.95, p=.016) after controlling for age (R^2=.50, $F(1,20)$=19.76, p<.001). See Appendix Tables 6-8 for results.

Comparison of children with low and high AHI

Using *t*-tests to compare low and high AHI groups, showed that findings on the SDQ were consistent with those of the hierarchical regression models; TD children with high AHI had increased conduct problems relative to those with low AHI (t(15=-3.29, p=.005, r=.65), decreased prosocial behaviour (t (15) = 2.49, p = .03, r = .54) and increased total problem score (t(15)=-2.63, p=.02, r=.56). In addition, there was a non-significant trend towards TD children with high AHI to have more problems with hyperactivity (t(15)=-1.98, p=.066, r=.46) (See Figure 1). We found no association between AHI and behaviour or cognition in the DS group, or on the Mullen Scales or MCDI for either group.

Figure 1

Discussion

Previous research indicates that children who experience sleep problems, including SDB, tend to show cognitive and behavioural deficits relative to children who sleep well (Karpinski et al.,

2008). In the current study, we used cardiorespiratory polygraphy alongside objective and parent-report measures to assess sleep, cognition and behaviour in TD children and children with DS.

As hypothesised, children with DS had increased SDB relative to TD children, although with greater inter-individual variability, ranging from normal sleep to severe OSAS. Children with DS were reported to sleep for one hour longer than TD children, driven by increased daytime sleep. In addition, as per hypothesis 2, children with DS exhibited lower scores on all objective and parent-reported cognitive scales, and they had more peer problems and less prosocial behaviour. In contrast, emotional symptoms, conduct problems, and hyperactivity were similar between groups, indicating areas of relative strength for children with DS.

Our third hypothesis was only partially supported: we found multiple associations between sleep, cognition and behaviour in TD children but not in children with DS. Notably, TD children with higher minimum SpO_2 had better expressive language skills, supporting previous research that SDB is related to verbal but not performance deficits (Blunden et al., 2005; Bourke et al., 2011). This finding is echoed in Breslin and colleague's (2014) data in children with DS, so it is possible that these findings could extend to DS given the right circumstances. Consistent with our hypotheses, TD children with fewer apnoeas demonstrated increased prosocial behaviour, and those with fewer apnoeas and hypopnoeas had reduced conduct problems and total problem score. Since SDB characteristics explained 11% to 29% of variance in scores, we are confident in confirming the relationship between SDB and cognitive and behavioural deficits in TD children. Although an effort was made to recruit children who snored, there was limited variability in SDB in TD children and none experienced severe sleep apnoea. We found that SDB at the mild end of the spectrum is sufficient to cognitively disadvantage children. This is noteworthy since mild sleep apnoea in children often goes unnoticed and there is no consensus on the precise severity at which it should be treated (Ahn, 2010; Marcus, 2010). Given that our findings support previous research, we suspect that they can extend to children with more extreme breathing difficulties, who might experience still

greater cognitive and behavioural deficits. It is therefore imperative that SDB is screened and treated from an early age, even at the mild end of the spectrum.

Additionally, we found an association between longer sleep duration and fewer emotional symptoms in TD, with sleep duration explaining 57% of the variance in scores after controlling for age. Sleep loss can be both a cause and a symptom of anxiety and other mood disturbances; however, treatment of sleep problems generally leads to an improvement in emotional symptoms in children (Dahl, 1996). We therefore propose that in the current study, shorter sleep duration is the cause of increased emotional problems, including fears, nervousness and unhappiness. Childhood sleep problems can predict anxiety disorders in adulthood (Gregory et al., 2005), so our finding of an association already evident in early childhood is particularly pertinent to children's future outcomes.

Longer sleep duration was also associated with increased parentally-reported expressive language on the MCDI. Interestingly, this does not echo the findings of our objective measure on the Mullen Scales, suggesting some difference between parent reports of language and children's performance on the day of testing.

That we did not find SDB to be associated with performance on the Mullen Scales or behaviour in children with DS, and our unexpected finding that increased Understanding and Actions and Gestures on the MCDI were associated with poorer SDB symptoms, might reflect that DS is a complex disorder where sleep is just one factor among many that might affect children's development. Alternatively, their longer sleep duration protected children with DS from the harmful effects of SDB on cognition. Lukowski and Milojevich (2016) showed that although parent-reported sleep problems were not correlated with temperament, mediation analysis revealed that sleep problems were associated with a greater increase in effortful and inhibitory control difficulties for preschoolers with DS, than for mental age-matched TD children; whereby sleep both mediated and was mediated by the association between group and temperament. We therefore expect that other factors which we did not control in the present study may have masked or mediated the effects of

sleep, for example, medical history, body mass index, previous hearing or vision problems, and

language difficulties mapping onto other areas; or that more sensitive cognitive measures may be

necessary to determine associations with sleep. It is also possible that difficulties experienced by

individuals with DS do not become apparent until later in childhood. Roberts and Richmond (2015)

demonstrated that although preschoolers with DS experience cognitive delay, they do not show the

characteristic pattern of weaknesses in memory and executive functions that are evident in

adolescents and adults with DS. Thus, it is possible that effects of SDB on certain cognitive domains

that have been demonstrated by other researchers (Andreou et al., 2002; Breslin et al., 2014) may not

yet be detectable in our young age group.

Tighter controls could account for the range of difficulties experienced by children with DS

as well as a larger sample size which might allow the detection of smaller effects. Indeed, following

the association between sleep and children's natural trajectory of development in longitudinal studies

would remove the need for tighter controls of confounding factors. Nevertheless, our sample of 22

children with DS was larger than many studies on sleep and in developmental disorders where

sample sizes are generally small (Andreou et al., 2002).

Limitations

We were not able to include snoring in the regression models because only around half of

our sample tolerated the nasal cannula. This is a particular problem when working with young

children, particularly those with intellectual disabilities who might have difficulties in understanding

the purpose of research. Despite problems tolerating equipment, we used an objective measure of

sleep because it is more reliable than parent report (Ashworth, Hill, Karmiloff-Smith, & Dimitriou,

2013; Shott et al., 2006) and because parents are unable to report on the respiratory symptoms of

interest.

In addition, it was not possible to gauge children's motivation to respond to instructions or

to maintain attention throughout the tasks. Indeed, parents frequently commented that their child was

capable of performing a particular task although they did not perform it during testing. The Mullen Scales take around one hour to administer and, despite breaks, maintaining attention was sometimes challenging. It would be beneficial for future research to maximise parent report alongside objective measures so that children's scores are not disadvantaged if they are unable to maintain performance on the test day.

Conclusions

In conclusion, our results support evidence that SDB in children is associated with poorer cognitive and behavioural functioning, particularly for expressive language, conduct problems and prosocial behaviour, whilst short sleep duration may lead to increased emotional problems and better parental reports of expressive language. Whilst we did find evidence of an association between worse SDB and parentally-reported better language understanding and use of actions and gestures in children with DS, we report these findings with the caveat that DS is a complex disorder and physiological and behavioural difficulties in these children may mask the effects of sleep disturbance on development. We recommend that all children be screened and treated for sleep problems since even at the mild end of the spectrum in a healthy population, they are sufficient to influence cognitive development; an important precursor to school readiness and future outcomes.

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Tables and Figures

Table 1

Participant details

	TD (*M* (*SE*))	DS (*M* (*SE*))	*t*	*p*	df	*r*
n	22	22				
Male/female ф	13/9	16/6	0.91	.53		.14
Age in months	39.03 (2.11)	36.57 (2.07)	0.83	.41	42	.13
Age range	25.79 – 59.53	24.38 – 56.48				
Gestational age*	39+6.55 (0.99)	38+6.41 (2.37)	-2.78	.01	28.14	.46
Maternal education (years)*	5.14 (0.72)	5.05 (0.48)	0.11	.92	36.50	.02
Paternal education (years)	3.82 (0.59)	3.55 (0.49)	.36	.72	42	.06

* Test statistics are adjusted for unequal variances.

ф Statistics are χ^2 (*t*) and Phi (*r*).

Table 2

Results of t-tests comparing TD and DS groups on Mullen Scales of Early Learning

	TD (*M* (*SE*)) *n* = 22	DS (*M* (*SE*)) *n* = 22	*t*	*p*	df	*r*
Gross Motor*	32.23 (.35)§	19.00 (1.22)	10.41	<.001	24.38	.90
Fine Motor*	40.22 (2.36)	19.50 (.87)	8.25	<.001	26.59	.85
Visual Reception*	48.77 (2.75)	22.95 (1.60)	8.10	<.001	33.76	.81
Receptive Language	45.86 (2.46)	22.82 (1.59)	7.86	<.001	42	.77
Expressive Language*	45.27 (2.77)	18.55 (1.16)	8.91	<.001	28.14	.86
Total raw score*	281.68 (21.08)	189.77 (14.18)	3.62	.001	36.79	.51

*Test statistics are adjusted for unequal variances.

§Note the maximum score is 33 for the Gross Motor scale and all except four children in the TD group performed at ceiling level for this scale.

Table 3

Results of t-tests comparing TD and DS groups on SDQ

	TD (*M* (*SE*))	DS (*M* (*SE*))	*t*	*p*	df	*r*
	n = 22	*n* = 22				
Emotional Symptoms	1.81 (0.38)	1.67 (0.50)	0.24	.81	41	.04
Conduct Problems	2.77 (0.40)	2.15 (0.29)	1.24	.22	40	.19
Hyperactivity	5.55 (0.47)	5.85 (0.62)	-0.39	.69	40	.06
Peer Problems	1.36 (0.41)	3.10 (0.45)	-2.86	.007	40	.41
Prosocial Behaviour	7.73 (0.43)	5.86 (0.43)	3.08	.004	41	.43
Total Problems	11.50 (1.17)	12.80 (1.28)	-0.75	.46	40	.12
Impact*	0.82 (0.28)	3.74 (0.80)	-3.02	.006	25.98	.51

*Test statistics are adjusted for unequal variances.

Table 4

Results of t-tests comparing TD and DS groups on MCDI

	TD (*M* (*SE*))	DS (*M* (*SE*))	*t*	*p*	df	*r*
	n = 15	*n* = 22				
Understands*	390.27 (10.60)	251.09 (21.29)	5.85	<.001	29.94	.73
Understands and Says	358.07 (15.30)	94.22 (21.12)	9.23	<.001	35	.84
Actions and Gestures*	56.07 (1.59)	46.73 (2.36)	3.28	.002	33.94	.49

*Test statistics are adjusted for unequal variances.

Table 5

Results of t-tests comparing TD and DS groups on cardiorespiratory polygraphy

	TD (*M* (*SE*))	DS (*M* (*SE*))	*t*	*p*	df	*r*
	n = 21	*n* = 22				
Recording duration (h:mm)	8:52 (0:40)	9:48 (0:26)	-1.18	.25	39	.19
Obstructive Apnoea Index	.53 (.26)	.99 (.59)	-.68	.50	39	.11
Apnoea Index	.91 (.29)	1.47 (.61)	-.79	.43	39	.13
Hypopnoea Index*	.39 (.11)	2.57 (1.02)	-2.13	.045	21.51	.42
Apnoea/Hypopnoea Index*	1.31 (.38)	4.03 (1.40)	-1.87	.07	24.04	.36
3% Desaturation Index*	1.64 (.42)	8.06 (2.20)	-2.86	.009	22.53	.52
Baseline SpO$_2$	98.79 (.20)	97.36 (.35)	3.39	.002	39	.48
Minimum SpO$_2$	90.89 (1.01)	88.36 (.90)	1.87	.07	39	.29
Night time sleep duration (hh:mm) ł	10:21 (0:18)	10:34 (0:10)	-0.62	.54	42	.10
Daytime sleep duration (hh:mm) ł	0:41 (0:10)	1:27 (0:10)	-3.19	.003	42	.44
Total sleep duration (hh:mm)* ł	11:03 (0:22)	12:02 (0.12)	-2.30	.03	32.42	.37

*Test statistics are adjusted for unequal variances.

ł Usual sleep duration was reported by parents and included both daytime and night time sleep.
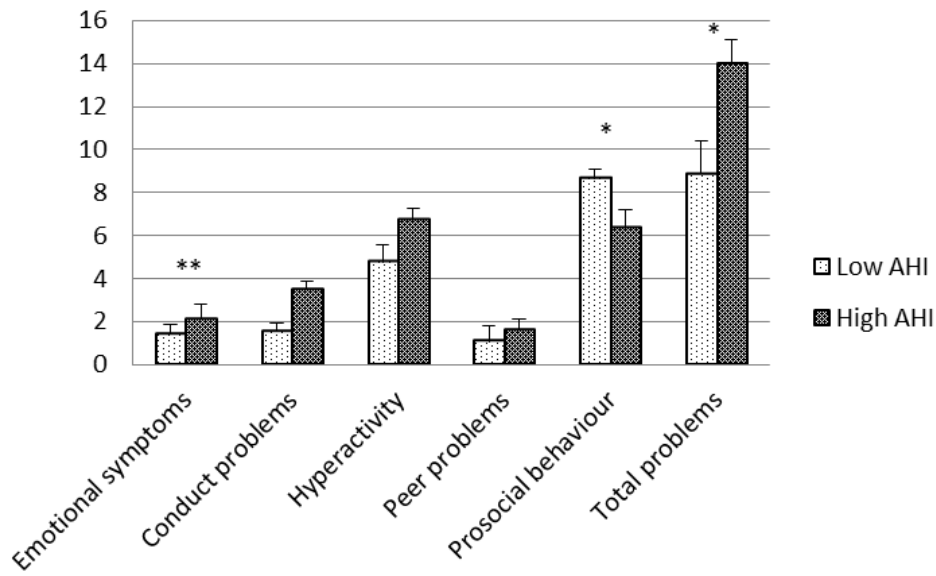
Figure 1. Comparison of typically developing children with high and low apnoea/hypopnoea index on the Strengths and Difficulties Questionnaire.

$* p < .05, ** p < .01.$

Appendix

Table 1
Hierarchical multiple regression results for TD children and children with DS for Expressive Language on Mullen Scales of Early Learning

				Overall model			Change statistics	
Group	Block	Predictors	*B*	*SE B*	*β*	ΔR^2	ΔF^2
TD	1	Constant	7.22	8.91		.55	21.12***
n = 19		Age (Months)	0.98	0.21	.74***		
	2	Constant	12.22	8.31		.11	5.04*
		Age (Months)	0.98	0.19	.74***		
		Sex	-8.40	3.74	-.33*		
	3	Constant	-82.27	35.03		.11	7.58***
		Age (Months)	1.04	0.16	.79***		
		Sex	-7.77	3.16	-.30*		
		Minimum SpO_2	1.01	0.37	.34*		
DS	1	Constant	5.40	3.64		.41	13.95
n = 22		Age (Months)	0.36	0.10	.64***		

*p < .05, *** p < .001.

Table 2
Hierarchical multiple regression results for TD children and children with DS for Emotional Problems on SDQ

				Overall model			Change statistics	
Group	Block	Predictors	*B*	*SE B*	*β*	ΔR^2	ΔF^2
TD	1	Constant	0.30	1.65		.04	0.69
n = 19		Age (Months)	0.03	0.04	.20		
	2	Constant	9.11	2.11		.57	23.68***
		Age (Months)	0.01	0.03	.04		
		Total sleep time	0.00	0.00	-.77***		
DS	1	Constant	-3.56	1.59		.38	11.54**
n = 21		Age (Months)	0.14	0.04	.61***		

** p < .01, *** p < .001.

Table 3
Hierarchical multiple regression results for Conduct Problems on SDQ

Group	Block	Predictors	Overall model			Change statistics	
			B	$SE\ B$	β	ΔR^2	ΔF^2
TD	1	Constant	1.02	1.54		.05	0.95
$n = 19$		Age (Months)	0.04	0.04	.23		
	2	Constant	0.29	1.35		.29	6.97*
		Age (Months)	0.04	0.03	.24		
		AHI	0.50	0.19	.54*		
DS	1	Constant	0.87	1.11		.07	1.44
$n = 20$		Age (Months)	0.04	0.03	.27		

* $p < .05$.

Table 4
Hierarchical multiple regression results for TD children and children with DS for the Prosocial scale on SDQ

Group	Block	Predictors	Overall model			Change statistics	
			B	$SE\ B$	β	ΔR^2	ΔF^2
TD	1	Constant	4.28	2.02		.16	3.19
$n = 19$		Age (Months)	0.09	0.05	.40		
	2	Constant	5.00	1.77		.25	6.80*
		Age (Months)	0.09	0.04	.40		
		Apnoea index	-0.84	0.32	-.50*		
DS	1	Constant	1.78	1.42		.32	8.88**
$n = 21$		Age (Months)	0.11	0.04	.56**		

* $p < .05$, ** $p < .01$.

Table 5
Hierarchical multiple regression results for TD children and children with DS for Total Problems on SDQ

Group	Block	Predictors	B	SE B	β	ΔR^2	ΔF^2
				Overall model		*Change statistics*	
TD	1	Constant	11.14	4.87		<.001	0.001
n = 17		Age (Months)	0.00	0.12	.01		
	2	Constant	9.26	4.43		.26	4.85*
		Age (Months)	0.00	0.11	.01		
		AHI	1.37	0.62	.51*		
DS	1	Constant	4.37	5.49		.15	2.71
n = 17		Age (Months)	0.23	0.14	.39		

* $p < .05$, ** $p < .01$, *** $p < .001$.

Table 6
Hierarchical multiple regression results for TD children and children with DS for the Understands scale on MCDI

Group	Block	Predictors	B	SE B	β	ΔR^2	ΔF^2
				Overall model		*Change statistics*	
TD	1	Constant	277.57	51.12		.35	5.42*
n = 12		Age (Months)	2.99	1.28	.59*		
DS	1	Constant	-28.36	58.43		.55	24.41***
n = 22		Age (Months)	7.64	1.55	.74***		
	2	Constant	2472.12	704.02		.18	12.67**
		Age (Months)	8.14	1.24	.79***		
		Baseline SpO_2	-25.87	7.27	-.43**		

* $p < .05$, ** $p < .01$, *** $p < .001$.

Table 7
Hierarchical multiple regression results for TD children and children with DS for the Understands and Says scale on MCDI

Group	Block	Predictors	Overall model			Change statistics	
			B	*SE B*	*β*	*ΔR²*	*ΔF²*
TD	1	Constant	205.86	61.32		.43	7.69*
n = 12		Age (Months)	4.28	1.54	.66*		
	2	Constant	26.24	87.25		.23	6.27*
		Age (Months)	4.31	1.25	.67**		
		Sleep duration	0.00	0.00	.48*		
DS	1	Constant	-105.53	73.06		.29	7.98**
n = 22		Age (Months)	5.46	1.93	.53**		

* p < .05, ** p < .01.

Table 8
Hierarchical multiple regression results for TD children and children with DS for the Actions and Gestures scale on MCDI

Group	Block	Predictors	Overall model			Change statistics	
			B	*SE B*	*β*	*ΔR²*	*ΔF²*
TD	1	Constant	44.89	8.90		.13	1.49
n = 12		Age (Months)	0.27	0.22	.36		
DS	1	Constant	17.29	6.84		.50	19.76***
n = 22		Age (Months)	0.80	0.18	.70***		
	2	Constant	10.63	6.51		.13	6.95*
		Age (Months)	0.90	0.16	.79***		
		Desaturation Index	0.40	0.15	.38*		

* p < .05, *** p < .001.