JIDR Journal of Intellectual Disability Research
Published on behalf of mencap and in association with IASSID

Journal of Intellectual Disability Research doi: 10.1111/jir.12321

VOLUME 61 PART 3 pp 221–232 MARCH 2017

Sleep problems and temperament in young children with Down syndrome and typically developing controls

A. F. Lukowski & H. M. Milojevich

Department of Psychology and Social Behavior, University of California-Irvine, Irvine, CA, USA

Abstract

Background Although group differences have been found between children with Down syndrome (DS) and typically developing (TD) children when considering sleep problems and temperament independently, none of the research conducted to date has examined sleep-temperament associations in children with DS. The present research was conducted to determine (1) whether the sleep problems experienced by children with DS are associated with temperament or (2) if the demonstrated relations between sleep and temperament differ from those that are observed in TD children.
Method The present study included examination of relations between parent-reported sleep problems and temperament in 19 children with DS and 20 TD controls matched on developmental age.
Results The results revealed group differences in temperament and sleep problems. Mediation models indicated that temperament (effortful control and inhibitory control) mediated the association between group and sleep problems; sleep problems also mediated the association between group and temperament (effortful and inhibitory control).
Conclusion Findings indicated that sleep problems may serve as both cause and consequence of variability in effortful and inhibitory control and provide insight as to future experimental studies that should be conducted to better elucidate these relations.

Keywords Down syndrome, effortful control, sleep, temperament, typically developing

Although children with Down syndrome (DS) commonly experience sleep problems (Carter *et al.* 2009; Breslin *et al.* 2011), there are limited data available to inform questions pertaining to (1) whether these sleep issues are associated with temperament or (2) if the demonstrated relations between sleep and temperament differ from those that are observed in typically developing (TD) children. The present study was conducted to address these questions through an examination of sleep-temperament relations in children with DS relative to a TD comparison group matched on developmental age (DA). Temperament was chosen as an indicator of daytime behaviour as work conducted with TD samples has revealed significant associations between sleep and temperament from infancy (for a review, see Ednick *et al.* 2009) to young adulthood (Lukowski & Milojevich 2014). As such, this literature provides a strong framework for evaluating and interpreting the findings obtained from children with DS. In addition, understanding associations between sleep and temperament in children with DS is practically important when considering the development or implementation of intervention efforts that might reduce sleep problems and facilitate optimal daytime functioning in children with DS.

Correspondence: Prof Angela F. Lukowski, Department of Psychology and Social Behavior, University of California-Irvine, 4304 Social and Behavioral Sciences Gateway, Irvine, 92697-7085 CA, USA (e-mail: angela.lukowski@uci.edu).

Sleep problems have been widely reported in children with DS, such that between 85% and 100% of school-aged children with DS experience clinically relevant levels of parent-reported sleep problems (Carter *et al.* 2009; Breslin *et al.* 2011). Sleep problems are multiply determined in children with DS, resulting from the same influences that impact sleep in TD children (Sadeh *et al.* 2010) as well as from additional unique challenges associated with the disorder. For example, children with DS oftentimes suffer from sleep-disordered breathing such as snoring or sleep apnea because of hypotonia, upper airway restriction and the placement of the tongue (for a review, see Churchill *et al.* 2012). Sleep-disordered breathing results in multiple brief sleep disruptions (sleep fragmentation) that may not wake the child but can reduce the restorative function of sleep and serve to increase daytime fatigue (Rosen 2011). Importantly, whereas sleep problems have been found to decline in TD infants and children over time, the same is not true for children with DS (Bartlett *et al.* 1985).

Although no research to our knowledge has examined relations between sleep and temperament in children with DS, a minority of studies has been conducted to examine whether sleep is associated with daytime behaviour in children with DS. One relevant study examined associations between parent-reported sleep problems and daytime behaviour in three groups of children: those with sleep onset issues, those with sleep maintenance issues and those with issues associated with sleep-disordered breathing (Stores *et al.* 1998). When compared with children with DS who did not have any sleep problems, those with sleep maintenance problems and those with issues associated with sleep-disordered breathing were more irritable and had more overall parent-reported behaviour problems; in addition, children without sleep problems were less hyperactive than children from the other three groups. Although not statistically significant in all cases, children with sleep maintenance problems had the most problematic daytime behaviour profiles. In another report, children with DS were more likely to snore, snore loudly and wheeze at night relative to their siblings without DS (Carskadon *et al.* 1993). Differences in parent-reported daytime behaviour were also apparent, such that children with DS were more likely to exhibit undesirable behaviours at school (including irritability, sleepiness and aggression). In addition, children with DS who were disruptive at school were more likely to snore at night, experience daytime sleepiness and take naps than those who were not identified as disruptive. Notably, associations between nighttime sleep and daytime functioning appear to extend to adolescence/young adulthood in those with DS, such that sleep problems were associated with reduced executive functioning, particularly verbal fluency and inhibitory control (Chen *et al.* 2013).

The literature examining sleep-behaviour associations in TD children has focused to a large extent on relations between sleep and temperament. Temperament has been defined as individual differences in reactivity and regulation that are due to genetic influences but are modifiable by environmental factors (Rothbart & Derryberry 1981). As reviewed previously (Gartstein *et al.* 2006), reactivity refers to the manner in which individuals react to environmental stimuli, whereas regulation refers to behavioural tactics that may be employed to modulate these responses. Work with TD infants indicates that night wakings or problematic sleep is associated with increased negative affect and reduced sensitivity to environmental stimuli (for a review, see Ednick *et al.* 2009); work with TD children and young adults also reveals associations between sleep problems and self-regulation, including increased impulsivity in adolescents (Moore *et al.* 2011) and reduced effortful control in young adults (Lukowski & Milojevich 2014).

Although a number of studies have been conducted to examine temperament in children with DS, the literature is largely inconclusive (Beeghly 1998) because of differences in the age of the participants, the inventories used to assess temperament and the group to which children with DS are compared. When temperament has been examined in children with DS relative to those who are TD, the most consistent finding is that children with DS are less persistent than those who are TD (Bridges & Cicchetti 1982; Gunn & Berry 1985a,1985b; Pueschel & Myers 1994). Additional findings indicate that 3-month-olds to 12-month-olds with DS experience higher levels of orienting/regulation (a factor characterised by positive ratings on cuddliness and soothability, among other temperament scales) and lower levels of negative affect (a factor including

higher ratings on sadness and fear, among other temperament scales) relative to TD infants matched on sex and chronological age (CA). Relative to their TD counterparts, infants with DS also experience less distress when placed in limiting situations, they obtain more pleasure from low-intensity stimuli (those that are relatively less intense, complex and novel than others), they attend to stimuli for greater durations of time, they are easier to comfort and they are cuddlier (Gartstein *et al*. 2006). Other studies indicate that infants with DS are more commonly identified as having difficult temperament profiles relative to controls matched on presumed DA, although this difference tends to diminish over time. As confirmation of this, 4-year-old to 11-year-old children with DS did not differ from TD controls matched on presumed DA when considering factor scores associated with effortful control (a factor including positive ratings on attention and inhibitory control, among other temperament scales), negative affect or surgency/extraversion (a factor characterised by higher ratings on activity level and impulsivity, among other temperament scales), although children with DS were rated as experiencing less attentional focusing, inhibitory control and sadness (Nygaard *et al*. 2002). Similarly, visual inspection of the group means reported in more recent research (Boström *et al*. 2010) indicated that children with DS were less emotional than TD children but were comparable on measures of parent-reported measures of activity, shyness, sociability and impulsivity (formal statistical analyses were not conducted comparing subgroups of children with intellectual disabilities to TD children).

Although group differences have been found between children with DS and TD children when considering sleep problems and temperament, none of the studies conducted to date have examined relations between sleep and temperament in children with DS when compared with a matched group of TD controls. Such research is important given practical implications associated with the development of intervention efforts that might reduce sleep problems and facilitate optimal daytime functioning in children with DS. To this end, the present study was conducted to address three aims. (1) The first aim is concerned whether children with DS experienced more sleep problems relative to their TD peers matched on DA. We expected that children with DS would experience more parent-reported sleep problems relative to TD children. If this prediction was realised, we would then examine which particular aspects of sleep were more problematic for children with DS relative to those who were TD. Based on previous research (Stores *et al*. 1998; Cotton & Richdale 2006; Breslin *et al*. 2011), we anticipated that relative to TD children, those with DS would experience more difficulty settling down to sleep at night, increased issues with sleep-disordered breathing and associated problems with daytime fatigue. (2) The second aim was to determine whether group differences were apparent in parent reports of child temperament. We predicted that our findings would most resemble those found previously (Nygaard *et al*. 2002) given the relative similarity in age between our respective participants (preschool to early childhood age) and the use of comparable measures of temperament (parent-report measures developed by Mary Rothbart and her colleagues). We expected that participants with DS would be rated lower on measures of effortful control relative to TD children, particularly when considering scales associated with attentional focusing and inhibitory control. (3) The third aim was to determine whether the associations between sleep and temperament differed for children with DS relative to those who were TD. To this end, we conducted mediation models to examine whether (1) sleep problems mediated the relation between group and temperament or (2) temperament mediated relations between group and sleep problems.

Method

Ethics

The study design and procedure were approved by the relevant state and university Institutional Review Boards. A waiver of written informed consent was granted for the questionnaire portion of the study, and parents signed informed consent statements at the first session.

Participants

Families were recruited to participate in a two-session study examining encoding and 1-month recall memory in children with DS and TD controls matched on DA (data from a subset of these children are presented in *blinded for review*). The present report

includes data from 19 children diagnosed with DS (mean age = 33 m 9d; range from 18 m 12d to 53 m 1d; 11 girls) and 20 TD children (mean age = 20 m 19d; range from 15 m 0d to 28 m 6d; 10 girls). An additional TD child was tested, but her scores were ultimately excluded from data analysis because of having a DA that was too high to match to a child with DS (DA = 36 months). Two additional children with DS were also tested but excluded from data analysis: one child experienced seizure activity shortly after birth, whereas the other child experienced significant prenatal and postnatal complications and was not developing as expected based on parent report.

Children with DS were recruited from local early intervention centres, organisations that provided educational and support services to children with DS and their families, and through snowball sampling. Families with TD children were initially contacted through a mass mailing about the possibility of participating in research studies on early memory development with their children. Interested families in both groups provided the research team with their phone number and email addresses and were contacted with additional information about participating in the study.

Demographic information is presented in Table 1. Of the 39 participants included in the final sample, 64% were Caucasian, 15% were Asian, 5% were African American and 13% were of mixed race; 3% of parents chose not to report race information. Twenty-six per cent of the children were of Hispanic ethnicity; the remaining children were non-Hispanic. Maternal education data were available for 96% of mothers. For those individuals, 79% had obtained at least a 4-year college degree. All parents received $30 in appreciation of their participation, and children received either a small toy or a junior scientist t-shirt.

Materials and measures

Questionnaires

Parents provided demographic information, including child race and ethnicity, and parental education, among other things. They also reported on the sleep habits of their children using the abbreviated version of the Children's Sleep Habits Questionnaire from the National Institute of Child Health and Human Development Study of Early Child Care and Youth Development (CSHQ-A; see also Goodlin-Jones *et al*. 2008; Owens *et al*. 2000; Seifer *et al*. 1996). Different versions of this questionnaire have been used previously to assess sleep problems in children with DS (Shott *et al*. 2006; Carter *et al*. 2009). In the present study, parents were asked to report how often their children experienced 22 sleep-related behaviours over the past week (1 = always or 7 days per week to 5 = never or 0 days per week). The questions were then combined as described later (Data Reduction: Questionnaires) to yield a sleep problems composite score. The questionnaire also asks parents to report on other measures of sleep behaviour, including weekday wake time, weekday bedtime, weekend wake time, weekend bedtime, nighttime sleep duration and daytime nap duration. One additional question was included as well to determine whether parents perceived the daytime and nighttime sleep behaviours of their children to be problematic (0 = not at all problematic to 4 = very problematic).

Parents reported on the temperament of their children using the short form of the Early Childhood Behaviour Questionnaire (Putnam *et al*. 2006). This 107-item questionnaire asks parents to report on child behaviour over the past week (1 = never to 7 = always). Parents could also indicate that their child did not

Table 1 Differences in demographic characteristics by group

	Down syndrome	Typically developing	Statistic	*P*-value	Effect size
Chronological age (months)*	33.29 ± 1.98	20.65 ± 1.93	$F = 20.96$	0.0001	$d = 1.45$
Developmental age (months)	21.63 ± 1.06	21.80 ± 1.03	$F = 0.01$	0.91	$d = 0.04$
Child sex (% girls)	58%	50%	$\chi^2 = 0.24$	0.62	$\phi = 0.62$
Child ethnicity (% Hispanic)	37%	15%	$\chi^2 = 2.44$	0.12	$\phi = 0.25$
Maternal education (% with at least a 4-year degree)	84%	90%	$\chi^2 = 0.29$	0.59	$\phi = 0.09$

Means and standard errors are presented for continuous variables.
*Significant differences between groups are indicated when $P < 0.05$.

experience the presented situation within the specified period of time.

Bayley Scales of Infant Development-III

The mental dimension of the Bayley Scales of Infant Development-III (BSID-III) was administered to all children at the first session for the purposes of matching groups on DA (Wright *et al.* 2006; Gilmore *et al.* 2009; Venuti *et al.* 2009; Reddy *et al.* 2010).

Procedure

Questionnaires

Parents received the questionnaires by mail along with a study information sheet that indicated that return of a completed questionnaire packet implied consent to participate.

Bayley Scales of Infant Development-III

The second author administered the mental portion of the BSID-III (Bayley 2006) to all children after informed consent was obtained at the first session.

Data reduction

Questionnaires

The data from the CSHQ-A were reduced to create a sleep problems composite. The composite was initially created by averaging the responses to the 22 items that inquired about the frequency with which children experienced sleep-related behaviours over the past week. The individual items were then correlated with the composite to identify the weakest correlations with $P \geq 0.10$. Of those associations, the questionnaire item that was least correlated with the sleep composite (in terms of the magnitude or absolute value of the association) was removed, and the composite score was recalculated. This procedure was repeated until all included items correlated with the sleep composite at $P \leq 0.10$. As shown in Table 2, the final sleep composite score included 19 items: 1, 2, 3, −4, −5, −7, −8, 10, −11, −12, −13, −14, −15, −17, −18, −20, −21 and −22 ($\alpha = 0.83$).

The responses to the individual items on the Early Childhood Behaviour Questionnaire were averaged to provide information on the 3 factor scores and 17 scales identified previously (Putnam *et al.* 2006). Factor scores included effortful control (individual scales pertained to attentional focusing, attentional shifting, cuddliness, inhibitory control and low-intensity pleasure), negative affect (individual scales pertained to discomfort, fear, frustration, motor activation, perceptual sensitivity, sadness, shyness and soothability) and extraversion or surgency (individual scales pertained to activity level, high-intensity pleasure, impulsivity, positive anticipation of pleasurable activities and sociability) (overall $\alpha = 0.90$).

Bayley Scales of Infant Development-III

The data obtained from the BSID-III were reduced as described in the administration manual (Bayley 2006). The second author matched the DAs of the children with DS to those who were TD within 3 months, as has been carried out in the previous research comparing these populations (Venuti *et al.* 2009). One TD child did not have a DA-matched peer with DS because of his exclusion for reasons mentioned earlier.

Results

Demographic information

Between-subjects analyses of variance (ANOVAs) were conducted by group (DS or TD) to determine whether the children with DS differed from those who were TD on any of the measured demographic characteristics. These data are shown in Table 1. Analyses revealed that the children with DS were chronologically older than the TD children but that the two groups did not differ on DA. As such, we were successful in our attempt to match the children across groups on DA.

We also conducted correlations separately by group to determine (1) whether CA was associated with DA or (2) whether CA and DA were associated with sleep problems. CA and DA were positively correlated in children with DS: $r(17) = 0.87$, $P < 0.0001$, as well as in TD children: $r(18) = 0.77$, $P < 0.0001$. In children with DS, sleep problems were unassociated with CA: $r(17) = 0.35$, $P = 0.15$ but were significantly related to DA: $r(17) = 0.47$, $P = 0.04$. In TD children, sleep problems were associated with neither CA: $r(18) = -0.09$, $P = 0.72$ nor DA: $r(18) = 0.03$, $P = 0.90$. Because CA was not differentially associated with DA by group and because CA was unrelated with sleep problems in children with DS

Table 2 Descriptive statistics and group differences on the Children's Sleep Habits Questionnaire-A (means and standard errors)

	Down syndrome	Typically developing	*F* statistic	*P*-value	*d* effect size
Sleep problems composite*	2.18 ± .12	1.83 ± .11	4.56	0.04	0.69
Q1. Child goes to bed at the same time each night.*	2.11 ± .10	1.80 ± .10	4.81	0.04	0.72
Q2. Child falls asleep within 20 min after going to bed.*	2.00 ± .15	1.53 ± .15	5.21	0.03	0.73
Q3. Child falls asleep alone in bed.	3.00 ± .35	2.10 ± .34	3.49	0.07	0.60
Q4. Child falls asleep in parent's or sibling's bed.	3.78 ± .35	4.22 ± .35	0.81	0.37	0.30
Q5. Child falls asleep with rocking or rhythmic movements.	3.84 ± .29	4.15 ± .28	0.58	0.45	0.25
Q7. Child needs parent in the room to fall asleep.	2.79 ± .40	3.42 ± .40	1.27	0.27	0.36
Q8. Child resists going to bed at bedtime.	3.42 ± .40	4.00 ± .24	2.97	0.09	0.56
Q10. Child sleeps the same amount each day.	2.00 ± .07	1.95 ± .06	—	—	—
Q11. Child is restless and moves much during sleep.	3.11 ± .21	3.56 ± .21	2.32	0.14	0.52
Q12. Child moves to someone else's bed during the night (parent, sibling etc.)	4.00 ± .25	4.45 ± .24	1.70	0.20	0.41
Q13. Child grinds teeth during sleep (your dentist may have told you this).	4.33 ± .19	4.78 ± .19	2.86	0.10	0.57
Q14. Child snores loudly.*	4.05 ± .15	4.55 ± .14	6.00	0.02	0.79
Q15. Child awakens during the night and is sweating, screaming and inconsolable.	4.79 ± .14	4.75 ± .13	0.04	0.84	0.07
Q17. Child wakes up once during the night.	3.95 ± .27	3.40 ± .26	2.09	0.16	0.46
Q18. Child wakes up more than once during the night.	4.37 ± .21	4.30 ± .20	0.06	0.82	0.08
Q20. Child wakes up very early in the morning (or, earlier than necessary or desired).	3.42 ± .26	3.60 ± .25	0.24	0.63	0.16
Q21. Child seems tired during the daytime.*	3.58 ± .13	4.15 ± .12	10.51	0.003	1.03
Q22. Child falls asleep while involved in activities.*	4.53 ± .11	4.95 ± .10	8.42	0.006	0.92
Additional sleep data					
Weekday wake time	6:40 ± :12	6:45 ± :12	0.11	0.75	0.10
Weeknight bedtime	20:25 ± :14	19:59 ± :14	1.69	0.20	0.42
Weekend wake time	6:55 ± :13	6:47 ± :13	0.20	0.66	0.14
Weekend bedtime	20:40 ± :16	20:07 ± :16	2.28	0.14	0.49
Night sleep duration*	701.05 ± 20.38	761.84 ± 20.38	4.45	0.04	0.68
Daytime nap duration	105.00 ± 7.97	98.95 ± 7.75	0.30	0.59	0.18
Parent perception of child sleep problems (nighttime and daytime combined)	0.74 ± 0.20	0.20 ± 0.19	3.87	0.06	0.63

Group differences could not be analysed for question 10 because there was no variability in the scores for children in the Down syndrome group (all participants received a score of 2). Although values on some of the individual questions were reverse scored for the purposes of creating the sleep composite, the unadjusted scores for the individual items are presented here (1 = always and 5 = never).
*Significant differences between groups are indicated when $P < 0.05$.

and TD children, CA is not included as a covariate in the subsequent analyses.

Sleep problems

Between-subjects ANOVAs were conducted by group (DS or TD) to determine whether children differed on parent-reported sleep habits. These data are shown in Table 2. Analyses indicated that children with DS experienced more sleep problems overall relative to children who were TD. Given this significant group difference, we examined on which individual components of sleep problems children with DS received higher scores. Relative to TD children, those with DS went to bed at the same time each night less frequently and more often required more than 20 min to fall asleep. Children with DS were more likely to snore loudly during sleep and obtained less sleep at night relative to children who were TD. In addition, children with DS were more likely to seem tired and fall asleep during daytime activities relative to children who were TD.

Temperament

Between-subjects ANOVAs were conducted by group (DS or TD) to determine whether children differed on parent-reported temperament. These data are shown in Table 3. Our analyses initially focused on the three factor scores pertaining to effortful control, negative affect and surgency. To reduce the number of comparisons made, we only examined whether group differences were found on the individual scales if group differences were obtained on the relevant higher order factor score.

The results revealed that children with DS received lower ratings on the factor pertaining to effortful control relative to children who were TD. Group differences were not found when considering negative affect or surgency. Given the significant group difference on effortful control, we examined on which individual scales children with DS received lower ratings. The analyses revealed that children with DS received lower scores on scales assessing attentional focusing, attentional shifting, cuddliness and inhibitory control relative to children who were TD; differences were not apparent on the scale assessing low-intensity pleasure.

Sleep-temperament associations

Correlations

We conducted correlations separately by group (DS or TD) to examine associations between sleep problems and temperament. Significant associations were not found for any of the analysed sleep-temperament associations: for children with DS, *r*s from −0.42 (effortful control) to 0.24 (surgency); for TD children, *r*s from −0.27 (effortful control) to 0.00 (negative affect and surgency).

Mediation analyses

Mediation analyses were conducted using the approach outlined in Hayes (2013) so as to examine relations between group, sleep problems and temperament. Because mediation models are used to specify a causal sequence of events, we specified group as the predictor in the conducted analyses as differences in developmental status most certainly precede differences in sleep problems and temperament instead of vice versa. Correct specification of the mediation and outcome variables was more tenuous; however, as the existing literature does not provide strong evidence to indicate whether variability in temperament precedes or results from differences in sleep problems. For this reason, we tested two alternate sets of mediation models (Fig. 1). Model 1 (panel A) examined whether the relation between group (0 = TD and 1 = DS) and temperament was mediated by sleep problems. Model 2 (panel B) examined an alternate model in which the relation between group (0 = TD and 1 = DS) and sleep problems was mediated by temperament. The proposed mediators (sleep problems in model 1 and temperament in model 2) were centred before analysis as 0 was not a meaningful value on either of them (in both cases, the minimum possible score was 1).

Mediation analyses were only conducted examining the effortful control factor score and the

Table 3 Descriptive statistics and group differences on the Early Childhood Behaviour Questionnaire (means and standard errors)

	Down syndrome (*n* = 19)	Typically developing (*n* = 20)	*F* statistic	*P*-value	*d* effect size
Effortful control*	4.23 ± .14	4.75 ± .13	7.51	0.009	0.89
Attentional focusing*	3.85 ± .19	4.57 ± .19	7.14	0.01	0.85
Attentional shifting*	4.47 ± .16	5.01 ± .16	5.76	0.02	0.75
Cuddliness*	4.61 ± .21	5.24 ± .21	4.50	0.04	0.67
Inhibitory control*	3.25 ± .23	4.16 ± .22	8.02	0.007	0.91
Low-intensity pleasure	4.95 ± .26	4.74 ± .25	0.36	0.55	0.19
Negative affect	2.84 ± .15	2.89 ± .15	0.06	0.81	0.08
Surgency	4.63 ± 0.16	4.86 ± 0.16	0.94	0.34	0.32

Average temperament scores ranged from 1–7, with higher scores indicating increased levels of the behaviour in question.
*Significant differences between groups are indicated when $P < 0.05$.

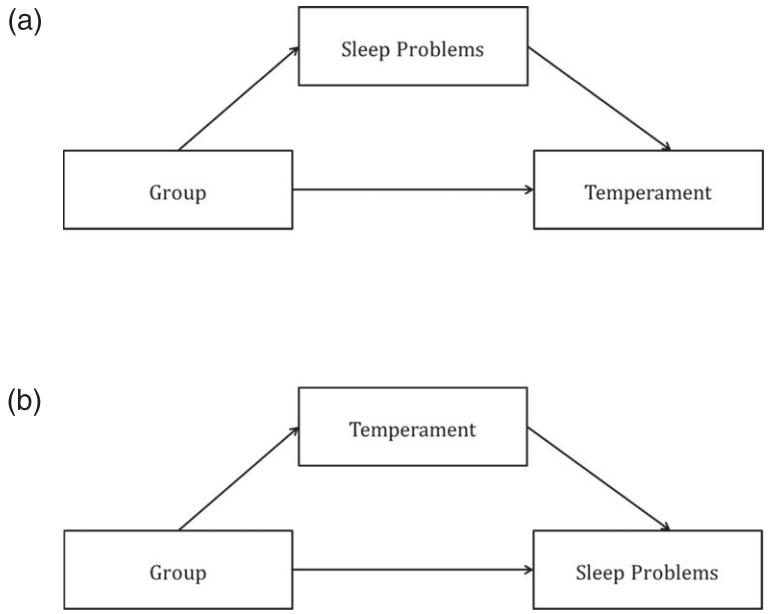

Figure 1 Mediation model 1 (A), in which sleep problems were predicted to mediate the association between group (0 = TD, 1 = DS) and temperament. Mediation model 2 (B), in which temperament was expected to mediate the association between group (0 = TD, 1 = DS) and sleep problems. Evidence of mediation was found when examining aspects of temperament associated with effortful control and inhibitory control; evidence of mediation was not obtained for temperament scales associated with attentional focusing, attentional shifting, cuddliness or low-intensity pleasure.

associated scale scores on which significant group differences were found. We chose to conduct mediation analyses only on these significant outcomes given (1) our goal of explaining associations that differed significantly by group as well as (2) to reduce the likelihood of obtaining significant results by chance (i.e. committing type I errors).

Effortful control

The analysis of model 1 revealed that group was positively associated with sleep problems ($a = 0.35$, $P = 0.04$) and that sleep problems were negatively associated with effortful control ($b = -0.40$, $P = 0.04$). Evidence of mediation was obtained from a bias-corrected bootstrap confidence interval that did not include 0 ($ab = -0.14$, confidence interval from -0.4426 to -0.0021). The direct effect of group on effortful control was not significant ($c' = -0.38$, $P = 0.05$).

The analysis of model 2 indicated that group was negatively associated with effortful control ($a = -0.52$, $P = 0.009$) and that effortful control was negatively associated with sleep problems ($b = -0.29$, $P = 0.04$). Evidence of mediation was obtained from a bias-corrected bootstrap confidence interval that did not include 0 ($ab = 0.15$, confidence interval from 0.0101 to 0.4085). The direct effect of group on sleep problems was not significant ($c' = 0.19$, $P = 0.26$).

Inhibitory control

The analysis of model 1 revealed that group was positively associated with sleep problems ($a = 0.35$, $P = 0.04$) but that sleep problems were unassociated with inhibitory control ($b = -0.57$, $P = 0.08$). Evidence of mediation was obtained from a bias-corrected bootstrap confidence interval that did not include 0 ($ab = -0.20$, confidence interval from -0.6564 to -0.0002). The direct effect of group on inhibitory control was not significant ($c' = -0.71$, $P = 0.04$).

The analysis of model 2 indicated that group was negatively associated with inhibitory control ($a = -0.90$, $P = 0.008$) and that inhibitory control was unrelated to sleep problems ($b = -0.15$, $P = 0.08$). Evidence of mediation was obtained from a bias-corrected bootstrap confidence interval that did not include 0 ($ab = 0.13$, confidence interval from 0.0053 to 0.3674). The direct effect of group on sleep problems was not significant ($c' = 0.21$, $P = 0.23$).

Other temperament scales

Evidence of mediation was not obtained in any of the tested models when considering attentional focusing, attentional shifting and cuddliness as mediators (model 1) or outcomes (model 2).

Discussion

Although associations between sleep and temperament have been studied for decades in TD infants and children (for a review, see Ednick *et al.* 2009), little research has been conducted to examine (1) whether sleep-temperament relations are found in children with developmental disabilities or (2) whether the observed associations differ from those observed for TD children. The presented research was conducted to address this gap in the literature.

Our hypotheses concerning group differences in sleep and temperament were largely supported. As expected, children with DS experienced more sleep problems relative to children who were TD. Additional examination of the individual questions that were used to create the sleep composite revealed that children with DS were less likely to go to bed at the same time each night and had longer sleep latencies relative to TD children. Children with DS also experienced increased snoring at night and greater evidence of daytime fatigue. Temperament profiles also differed by group. As predicted, children with DS were rated as experiencing less effortful control relative to their TD peers matched on DA. Examination of the individual scales that comprised the effortful control factor score revealed that children with DS received lower ratings on attentional focusing, attentional shifting, cuddliness and inhibitory control relative to TD children (differences were not found on the scale pertaining to low-intensity pleasure).

The primary contribution of the present research, however, is in our use of mediation models to determine (1) whether sleep problems mediated the relation between group and temperament and/or (2) whether temperament mediated the relation between group and sleep problems. The results obtained from model 1 revealed that at an average level of sleep problems (due to the mean-centring procedure), children with DS experienced reduced effortful control and inhibitory control compared with TD children. In a complementary fashion, the findings from model 2 indicated that at average levels of effortful control and inhibitory control (again due to the mean-centring procedures), children with DS experienced increased sleep problems compared with TD children. Importantly, however, different patterns of significant associations were obtained within models when examining effortful control and inhibitory control. For example, when group was held constant in model 1, mean sleep problems predicted variability in effortful control but not inhibitory control. Similarly, when group was held constant in model 2, average levels of effortful control – but not inhibitory control – predicted sleep problems. These findings indicate that the included measures of effortful and inhibitory control are in some ways distinct, although the inhibitory control temperament scale is contained with the factor score pertaining to effortful control.

Taken together, the presented results suggest that sleep problems may serve as both cause and consequence of variability in effortful control and inhibitory control in children with DS relative to those who are TD. Given that this conclusion was drawn using statistical models of causal relations in the absence of an experimental design, one significant direction for future research involves experimental examination of potential interventions that explicitly address the dual role sleep problems may play in effortful and inhibitory control processes (Dowsett & Livesey 2000; Thorell *et al.* 2009). Importantly, these findings were obtained from a sample that is not likely biased towards children with sleep problems, as families were recruited to participate in a study examining recall memory in children with DS and TD children matched on DA. Future research should similarly ensure that the recruited samples are not biased to families who are experiencing child sleep problems. Subsequent studies should also include objective assessments of sleep and temperament, as the reported findings were obtained only through parent-report questionnaires and previous research has documented unique sleep-temperament associations when sleep data are recorded subjectively and objectively (Scher *et al.* 1992).

Future work should also attempt to overcome the limitations of the present study. Although comparable in size to other published research on children with DS ($n = 17$, as in Gartstein *et al.* 2006; $n = 15$, as in Gunn *et al.* 1983), subsequent research should include a greater number of participants. Future researchers should also include a more comprehensive sleep questionnaire than the CSHQ-A. We used this questionnaire because of its ability to assess sleep problems instead of variability in usual sleep habits using a relatively small number of items. However, this questionnaire is limited in that we were unable to

determine whether issues with sleep-disordered breathing or behavioural sleep problems differentially contributed to variability in parent-reported temperament. This shortcoming is noteworthy, as children with DS commonly experience issues associated with sleep apnea and sleep-disordered breathing (Carskadon *et al.* 1993; Carter *et al.* 2009; Breslin *et al.* 2011). Future research should address this issue by using the full-scale CSHQ (Owens *et al.* 2000), which includes scales that assess various sleep problems (such as bedtime resistance, sleep onset delay, sleep anxiety, night wakings, sleep-disordered breathing and daytime sleepiness, among others) and has been validated for use with toddlers and preschoolers as well as children with developmental delays (autism and developmental delay in the absence of autism; Goodlin-Jones *et al.* 2008). Parents should also be asked to reflect separately on perceived problems with child daytime and nighttime sleep habits, as previous research has indicated that children with DS experience significant daytime sleepiness (Carter *et al.* 2009; Breslin *et al.* 2011) that may impair child functioning.

In addition to these procedural modifications, future research should also be conducted to help explain some of the discrepant findings across studies. As we noted in the introduction, study-related variability exists in the age of the participants, the inventories used to assess temperament and the groups to which children with DS are compared. These methodological differences are likely meaningful, for example, the results of this research indicated that parents rated children with DS as less cuddly relative to TD children matched on DA, whereas Gartstein *et al.* (2006) noted that parents rated infants with DS as cuddlier than TD infants matched on CA. We matched children on DA in the present research because the primary goal of the study into which they were recruited was to examine group differences in encoding and 1-month-delayed recall memory, and matching on DA relative to CA is relatively common in this literature (e.g. Gilmore *et al.* 2009; Roberts & Richmond 2015, among others). Although the reported variability in CA could have contributed to parent-reported sleep problems in our sample, CA was unrelated to sleep problems in both groups of children. In addition, CA was significantly correlated with DA in each group of participants. For these reasons, we did not statistically adjust for CA in the conducted analyses. However, future research should adopt a more nuanced approach to understanding associations among CA, DA and sleep-temperament associations by testing children with DS in addition to two TD control groups (one matched on CA and another matched on DA).

Although additional work remains to be done, the conducted study represents an important first step in examining possible causal relations between sleep and temperament in children with DS relative to those who are TD. Findings indicated that sleep problems may serve as both cause and consequence of variability in effortful and inhibitory control and provide insight as to future experimental studies that should be conducted to better elucidate these relations. The completion of such research will provide empirical evidence as to which behaviours to manipulate in intervention efforts, with the ultimate goal of reducing sleep problems and facilitating optimal daytime functioning in children with DS.

References

Bartlett B., Rooney V. & Spedding S. (1985) Noctural difficulties in a population of mentally handicapped children. *The British Journal of Mental Subnormality* **31**, 54–9.

Bayley N. (2006) *Bayley Scales of Infant and Toddler Development-III*. Harcourt Assessment, San Antonio, TX.

Beeghly M. (1998) Temperament in children with Down syndrome. In: *What Do We Know about Down Syndrome: A Review of Current Knowledge* (eds J. A. Rondal, J. Perera & L. Nadel), pp. 111–23. C. Whurr, London.

Boström P., Broberg M. & Hwang C. P. (2010) Different, difficult or distinct? Mothers' and fathers' perceptions of temperament in children with and without intellectual disabilities. *Journal of Intellectual Disability Research* **54**, 806–19.

Breslin J. H., Edgin J. O., Bootzin R. R., Goodwin J. L. & Nadel L. (2011) Parental report of sleep problems in Down syndrome. *Journal of Intellectual Disability Research* **55**, 1086–91.

Bridges F. A. & Cicchetti D. (1982) Mothers' ratings of the temperament characteristics of Down syndrome infants. *Developmental Psychology* **18**, 238–44.

Carskadon M. A., Pueschel S. M. & Millman R. P. (1993) Sleep-disordered breathing and behavior in three risk groups: preliminary findings from parental reports. *Child's Nervous System*, 452–7.

Carter M., McCaughey E., Annaz D. & Hill C. M. (2009) Sleep problems in a Down syndrome population. *Archives of Disease in Childhood* **94**, 308–10.

Chen C. C., Spanò G. & Edgin J. O. (2013) The impact of sleep disruption on executive function in Down syndrome. *Research in Developmental Disabilities* **34**, 2033–9.

Churchill S. S., Kieckhefer G. M., Landis C. A. & Ward T. M. (2012) Sleep measurement and monitoring in children with Down syndrome: a review of the literature, 1960–2010. *Sleep Medicine Reviews* **16**, 477–88.

Cotton S. & Richdale A. (2006) Parental descriptions of sleep problems in children with autism, Down syndrome, and Prader–Willi syndrome. *Research in Developmental Disabilities* **27**, 151–61.

Dowsett S. M. & Livesey D. J. (2000) The development of inhibitory control in preschool children: effects of "executive skills" training. *Developmental Psychobiology* **36**, 161–74.

Ednick M., Cohen A. P., McPhail G. L., Beebe D., Simakajornboon N., Amin R. S. *et al.* (2009) A review of the effects of sleep during the first year of life on cognitive, psychomotor, and temperament development. *Sleep* **32**, 1449–58.

Gartstein M. A., Marmion J. & Swanson H. L. (2006) Infant temperament: an evaluation of children with Down syndrome. *Journal of Reproductive and Infant Psychology* **24**, 31–41.

Gilmore L., Cuskelly M., Jobling A. & Hayes A. (2009) Maternal support for autonomy: relationships with persistence for children with Down syndrome and typically developing children. *Research in Developmental Disabilities* **30**, 1023–33.

Goodlin-Jones B. L., Sitnick S. L., Tang K. & Liu J. (2008) The Children's Sleep Habits Questionnaires in toddlers and preschool children. *Journal of Developmental and Behavioral Pediatrics* **29**, 82–8.

Gunn P. & Berry P. (1985a) Down's syndrome temperament and maternal response to descriptions of child behavior. *Developmental Psychology* **21**, 842–7.

Gunn P. & Berry P. (1985b) The temperament of Down's syndrome toddlers and their siblings. *Journal of Child Psychology and Psychiatry* **26**, 973–9.

Gunn P., Berry P. & Andrews R. J. (1983) The temperament of Down's syndrome infants: a research note. *Developmental Psychology* **22**, 189–94.

Hayes A. F. (2013) *Introduction to mediation, moderation, and conditional process analysis: a regression-based approach*. The Guilford Press, New York.

Lukowski A. F. & Milojevich H. M. (2014) Sleep quality and temperament among university students: differential associations with nighttime sleep duration and disruptions. *Behavioral Sleep Medicine* **12**, 1–14.

Marcovitch S., Goldberg S., MacGregor D. L. & Lojkasek M. (1986) Patterns of temperament variation in three groups of developmentally delayed preschool children: Mother and father ratings. *Developmental and Behavioral Pediatrics* **7**, 247–52.

Moore M., Slane J., Mindell J. A., Burt S. A. & Klump K. L. (2011) Sleep problems and temperament in adolescents. *Child: Care, Health and Development* **37**, 559–62.

Nygaard E., Smith L. & Torgersen A. M. (2002) Temperament in children with Down syndrome and in prematurely born children. *Scandinavian Journal of Psychology* **43**, 61–71.

Owens J. A., Spirito A. & McGuinn M. (2000) The Children's Sleep Habits Questionnaire (CSHQ): psychometric properties of a survey instrument for school-aged children. *Sleep* **23**, 1043–51.

Pueschel S. M. & Myers B. A. (1994) Environmental and temperament assessments of children with Down's syndrome. *Journal of Intellectual Disability Research* **38**, 195–202.

Putnam S. P., Gartstein M. A. & Rothbart M. K. (2006) Measurement of fine-grained aspects of toddler temperament: The Early Childhood Behavior Questionnaire. *Infant Behavior and Development* **29**, 386–401.

Reddy V., Williams E., Costantini C. & Lan B. (2010) Engaging with the self: mirror behaviour in autism, Down syndrome and typical development. *Autism* **14**, 531–46.

Roberts L. V. & Richmond J. L. (2015) Preschoolers with Down syndrome do not yet show the learning and memory impairments seen in adults with Down syndrome. *Developmental Science* **18**, 404–19.

Rosen D. (2011) Management of obstructive sleep apnea associated with Down syndrome and other craniofacial dysmorphologies. *Current Opinion in Pulmonary Medicine* **17**, 431–6.

Rothbart M. K. & Derryberry D. (1981) Development of individual differences in temperament. In: *Advances in Developmental Psychology* (eds M. E. Lamb & A. L. Brown), pp. 37–86. Lawrence Erlbaum, Hillsdale.

Sadeh A., Tikotzky L. & Scher A. (2010) Parenting and infant sleep. *Sleep Medicine Reviews* **14**, 89–96.

Scher A., Epstein R., Sadeh A., Tirosh E. & Lavie P. (1992) Toddlers' sleep and temperament: reporting bias or a valid link? *Journal of Child Psychology and Psychiatry, and Allied Disciplines* **33**, 1249–54.

Seifer R., Sameroff A. J., Dickstein S. & Hayden L. C. (1996) Parental psychopathology and sleep variation in children. *Child and Adolescent Psychiatric Clinics of North America* **5**, 715–27.

Shott S. R., Amin R., Chini B., Heubi C., Hotze S., Akers R. *et al.* (2006) Obstructive sleep apnea: should all children with Down syndrome be tested? *Archives of Otolaryngology - Head and Neck Surgery* **132**, 432–6.

Stores R., Stores G., Fellows B. & Buckley S. (1998) A factor analysis of sleep problems and their psychological associations in children with Down's syndrome. *Journal of Applied Research in Intellectual Disabilities* **11**, 345–54.

Thorell L. B., Lindqvist S., Bergman Nutley S., Bohlin G. & Klingberg T. (2009) Training and transfer effects of executive functions in preschool children. *Developmental Science* **12**, 106–13.

Venuti P., de Falco S., Esposito G. & Bornstein M. H. (2009) Mother-child play: children with Down syndrome and typical development. *American Journal on Intellectual and Developmental Disabilities* **114**, 274–88.

Wright I., Lewis V. & Collis G. M. (2006) Imitation and representational development in young children with Down syndrome. *Journal of Speech, Language, and Hearing Research* **56**, 429–50.

Accepted 29 June 2016