Original Article

Sleep in infants and toddlers with Down syndrome compared to typically developing peers: looking beyond snoring

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ABSTRACT

Aims: To compare sleep in infants and toddlers with Down syndrome (DS) to typically developing controls, including differences in snoring and sleep ecology (sleep setting and parent behaviors).
Methods: Parents of 104 children with DS and 489 controls aged 6–36 months completed the Brief Infant Sleep Questionnaire (BISQ). We explored group differences, controlling for demographic variables.
Results: Parents of children with DS reported more sleep problems (45% v 19%), snoring (19% vs 2%), room-sharing (37% vs 17%), as well as less night-time sleep (55 mins) and total sleep over 24 h (38 mins). They were more likely to be present when their child fell asleep (OR 4.40). Snoring increased night waking but did not limit night-time/24-hour sleep. However, parental presence was associated with 55 min less night-time and 64 min less 24-hour sleep. After controlling for snoring and parental presence, children with DS slept less at night (38 mins) but more during the day (21 mins) with no significant difference in 24-hour sleep.
Conclusions: Overall, significant differences in sleep patterns, problems, and ecology were found between children with DS and controls. Parental presence at settling, not snoring, explained most differences, including over an hour's less 24-hour sleep. Early intervention programmes that promote self-soothing skills could prevent the burden of sleep loss in young children with DS.

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1. Background

Down syndrome (DS), caused by partial or complete trisomy of chromosome 21, occurs in approximately 1:1200 live births worldwide [1]. Notably, it accounts for 12–15% of all individuals with learning disabilities in developed countries [2]. While the medical comorbidities of DS (eg, hypothyroidism and sensory impairments) are managed within medical surveillance and treatment programmes [3], sleep problems remain a hidden epidemic, affecting up to 85% of school-aged children with DS [4]. The most widely reported sleep disorder in DS is obstructive sleep apnea with prevalence rates around 75% [5], compared to 1–3% in the typically developing (TD) child population. While this is an important focus of practice and research, it may not fully explain the sleep problems reported by parents. Children with DS are children first and will also be vulnerable to behavioral insomnia [6]. Behavioral insomnias arise when children fail to learn the art of self-soothing to sleep. There are two core types [7]. First, 'sleep onset association disorder' - which typically arises when a parent is present as the child falls asleep at

bedtime. Settling may be rapid and mutually rewarding, but the child struggles to self-soothe after natural night wakings unless the parent is present. This is perceived as 'troublesome' night waking. Second, limit-setting behavioral insomnia when parents struggle to enforce boundaries and limits at bedtime, resulting in difficulties settling to sleep. These disorders can be effectively treated through modification of parent behaviors [8] to teach self-soothing skills alongside appropriate sleep hygiene approaches [9,10]. Prompt treatment of sleep disorders is necessary as poor quality sleep is associated with a wide range of adverse outcomes in childhood including obesity [11,12], cognitive and behavioral problems [13], and poor school performance [14]. Moreover, exposure to sleep problems in the early years of life may impact neurodevelopment as sleep may facilitate functional maturation of the brain [15]. This is supported by large longitudinal studies showing associations between sleep problems at two years, cognitive difficulties later in childhood [16] and reduced grey matter volume [17].

Few studies have reported sleep problems in infants and toddlers (less than three years of age) with DS [18—23]. In an online survey of 253 parents of children with DS aged 0—18 years in the US [19] 33% reported that their child frequently, or almost always, had difficulties initiating or maintaining sleep; moreover, 51% were concerned about sleep disordered breathing. While this study included approximately 68 infants and toddlers, data were not presented for those aged 0—3 years separately. Four further studies included infant and toddler data [18,20,21,23], in three, sample size was less than 20, none included a control group, and all used the Children's Sleep Habits questionnaire, which is not validated for this age group. Despite these limitations, studies consistently identified problems initiating sleep, restless sleep, night wakings and symptoms of sleep disordered breathing. Fernandez and colleagues objectively measured sleep for one week with continuous actigraphy in 66 children with DS aged 5—67 months and 43 TD age-matched controls. One third of the DS children were phase advanced, with earlier morning waking, and children with DS had lower sleep efficiency (75.9% v 82.9%) and shorter total sleep times than typically developing (TD) controls [22].

In summary, infants and toddlers with DS have been consistently reported to have poor sleep quality and symptoms of sleep disordered breathing. While it is reasonable to assume that sleep disordered breathing is a cause of sleep disruption, there are no existing data examining the role of sleep ecology (sleep setting and parent behaviors) in this population.

In the present study, we aimed to explore in detail sleep patterns and sleep ecology in infants and toddlers with DS compared to a TD control group. Furthermore, we aimed to explore the extent to which a child's snoring status and parental behaviors (specifically presence at bedtime) explained any differences in sleep outcomes between the DS and TD groups.

2. Methods

2.1. Participants

2.1.1. Down syndrome

Recruitment of children aged six months to six years with DS took place between 2012 and 2015 in three sites in the UK (Sheffield, Evelina London and Southampton Children's Hospitals) in a study of obstructive sleep apnea syndrome [24]. Children receiving home oxygen or non-invasive ventilation therapy were excluded. Multiple recruitment routes were used to increase study generalisability including child development centres, local children's hospitals, DS support groups and parent word of mouth. Of 202 children recruited, 104 children were aged six to 36 months and included here.

2.1.2. Typically developing controls

UK controls were sourced from an international internet-based survey of children's sleep habits conducted between May and June 2006 [25]. The online questionnaire was displayed as a pop-up screen on a parenting website (BabyCentre), inviting parents to complete a sleep survey for children aged from birth to 36 months. A total of 800 questionnaires were completed, of which 586 related to children aged six to 36 months. Of these, 82 were excluded due to a reported health problem in the child and 15 due to missing demographic data. Analysis was based on the remaining 489 questionnaires.

2.2. Measures

2.2.1. Demographic information

Parents reported basic demographic information including their highest educational level, child's gender, age in months (DS) and age category: 6—8, 9—11, 12—17, 18—23 and 24—36 months (TD group). DS data were re-coded to match the TD age categories and the categories 6—8 months and 9—11 months were combined to a single category: 6—11 months.

2.2.2. Snoring status

Parents of children with DS were asked: 'How often does your child snore when they do not have a cold?' with response options: Never, Rarely, Occasionally, Almost Always, Always. Parents of TD children were asked: 'Does your child snore during sleep?' with response options: Never, Only when he/she has a cold or allergy, Sometimes, Always or Almost Always. A new variable was generated 'Does the child snore (almost always/always)?' and coded 'Yes' for responses 'Almost always and always' and 'No' for all other possible responses: Never, Rarely, Occasionally, Sometimes, Only when he/she has a cold or allergy.

2.2.3. Sleep habits

An extended version of the Brief Infant Sleep Questionnaire (BISQ) was completed by parents of both groups. For children with DS, questionnaires were paper based. The BISQ has been validated against objective actigraphy sleep measures and sleep logs with significant correlations between sleep onset time (p < 0.001 and p < 0.001 respectively); sleep duration (p < 0.05 for both) and night wakings (p < 0.0001 for both) [26]. It comprises 28 questions covering both sleep patterns (scheduling of night-time sleep and day-time naps) and sleep behaviors including multi-response options for location of sleep, settling routines and parental perception of child's sleep. Total 24-hour sleep time was calculated by summing total time asleep at night and total time asleep during the day. A new variable 'parental presence at bedtime' was computed from responses to: 'How does your child fall asleep most of the time?'. A similar variable 'parental presence during waking' was created in relation to response to: 'When your child wakes up during the night, what do you do?'. For example, when parents reported bottle feeding/rocking/holding their child this was coded as 'parental presence at bedtime' and when parents reported that the child settled alone in their own crib/bed or that after night waking they allowed their child to 'cry and fall back to sleep by himself/herself' this was coded as parental absence. Where responses were unclear, for example a child reported as settling to sleep while watching television (TV), this was coded as indeterminate and excluded from analysis.

2.3. Governance approvals and informed consent

For children with DS, parents provided informed consent. Ethics permissions were granted by the UK National Research Ethics

Committee (reference number 13/SC/0106). The study of TD children was approved by the Institutional Review Board at Saint Joseph's University, USA.

2.4. Statistical analysis

The distributions of gender, age, educational level and snoring status were described in each group and tested uncontrolled using Pearson's chi-squared test or the Mann Whitney U test as appropriate. Time variables were treated as continuous and means from the DS and TD groups compared in linear regression including three sets of controlling variables: model 1 — gender and factors for age and educational level; model 2 — model 1 variables plus snoring yes/no; and model 3 — model 2 variables plus whether, or not, the parent was present at bedtime. Differences in means and 95% confidence intervals (95% CI) will be presented for the DS-TD, snoring-not, and parent present-absent comparisons along with their significance from F tests of removing each term from the specified model. Means in the DS and TD groups adjusted for the covariates in model 1 will be presented. DS and TD groups were compared with respect to binary dependent variables in logistic regression controlling for covariates in models 1, 2 or 3 above, or for model 1 covariates plus parental presence at bedtime. Odds ratios (OR) and 95% CIs will be presented for DS/TD, snoring/not, and parent present/absent comparisons, along with their significance from likelihood ratio tests of removing each term from the model. Multiple response outcomes were first dichotomised and then analysed using binary logistic regression as described above. Data were analysed in SPSS v25.

3. Results

3.1. Demographic data and snoring status

DS and TD groups were similar with respect to gender, but children with DS were older and their main caregiver had lower educational attainment than in the control group (Table 1). As would be expected, snoring always/almost always was significantly more common in children with DS (19%) than TD controls (2%).

3.2. Sleep location

Across the sample, the majority (61%) of children slept in their own bedrooms (Table 2). Children with DS (37%) were more likely to sleep in their parents' bedroom compared to controls (17%); with an OR of 3.28 after controlling for demographic variables. Overall co-sleeping in the parental bed was rare, reported in 8% of the DS groups and 5% of the TD group.

3.3. Bedtime-routines

After controlling for demographic variables (Table 3), parents of children with DS were more likely than those of TD children to rock (OR 4.61) and massage (OR 2.32) their child, sing songs or say prayers in the hour before bedtime, but were less likely to bathe their child (OR 0.43) or read books to them (OR 0.46). The groups were similar with respect to both breast or bottle feeding during this hour, an activity that most parents reported (80%); having dinner or a snack (reported by 10%), running around (reported by 13%), listening to music (reported by 18%), and TV watching (reported by 24%). Overall, 51% of parents reported the same bedtime routine every night of the week, with no significant difference between groups (Table 4). Parents who were present in the room when their child fell asleep were more likely to report that they stuck to the same routine every night (OR 2.31).

3.4. Settling to sleep

After controlling for demographic variables, children with DS were significantly more likely to fall asleep while bottle feeding (OR 4.75), being held (OR 5.92) or rocked (OR 5.87) compared to TD peers (Table 3). Only 10% of all mothers breast-fed their child to sleep with no difference between the groups in this activity. Whilst most (72%) fell asleep alone in their own cot or bed, children with DS were more likely to fall asleep with a parent present, either in their own cot/bed (OR 2.04) or in the parental bed (OR 3.60). Parental presence to help the child settle to sleep was more likely in children with DS than controls (56% v 25%) with an OR of 4.40 after controlling for demographic variables. Few children fell asleep whilst watching TV (3.2%) with no differences between the groups. Despite differences in practices settling children to sleep, parents in both groups reported similar levels of ease or difficulty at bedtime (Table 4). Parents present as their child fell asleep were less likely to report that their children took longer than 15 min to fall asleep (OR 0.30) or to report bedtime as difficult (OR 0.19). Parental presence appeared to be associated with fewer difficulties in initially settling children to sleep.

3.5. Parental management of night wakings

After controlling for demographic variables, parents of children with DS were more likely to engage in active techniques to settle their child back to sleep after night waking such as rocking (OR 6.41); singing (OR 2.52), bottle feeding (OR 2.21), offering a dummy/pacifier (2.16), or bringing them into the parental bed (OR 2.31), see Table 5. TV/DVD use during the night was only reported by four parents. Overall, parents of children with DS were

Table 1
Characteristics of the groups.

		DS (n = 104)	TD (n = 489)	P
Gender	male	52 (50%)	221 (45%)	0.372[a]
	female	52 (50%)	268 (55%)	
Age in months	6 to 11	28 (27%)	199 (41%)	0.000[b]
	12 to 17	18 (17%)	100 (20%)	
	18 to 23	21 (20%)	93 (19%)	
	24 to 36	37 (36%)	97 (20%)	
Education of primary care giver	degree or higher	43 (41%)	289 (59%)	0.000[b]
	A level	18 (17%)	115 (24%)	
	below A level	43 (41%)	85 (17%)	
Almost always/always snores at night		30/103 (19%)	8/489 (2%)	0.000[a]

[a] Pearson's chi-squared test.
[b] Mann Whitney U test.

Table 2
Usual sleep location.

		Number (%)		MODEL 1 group, age, gender, educational level	
		DS (n = 104)	TD (n = 489)	DS/TD OR (95%CI)	P[a]
Where does your child sleep most of the time?	own bedroom	60 (58%)	371 (76%)		
	bedroom with sibs	4 (4%)	32 (7%)		
	other	2 (2%)	2 (0%)		
	parent's bedroom	38 (37%)	84 (17%)	3.28 (1.97, 5.47)	0.000
What does your child sleep in most of the time?	cot/crib	86 (83%)	316 (65%)		
	bassinet	2 (2%)	7 (1.4%)		
	own Bed	6 (6%)	142 (29%)		
	other	2 (2%)	0		
	parent's bed	8 (8%)	24 (5%)	1.23 (0.51, 2.97)	0.649

[a] Likelihood ratio test for removal of DS/TD term from model 1.

significantly more likely to stay with their child until they had settled back to sleep than parents of TD children, (74% v 56%) with an OR of 2.45 after controlling for demographic variables.

3.6. Sleep patterns

Night-time sleep patterns of children with DS were significantly worse in all domains than in TD children after adjusting for demographic variables (Tables 6 and 7, model 1). On average, they went to bed just over half an hour later; were more likely to wake more than once during the night (36% v 21%) with an OR 2.48; experienced an uninterrupted sleep period that was on average 85 min shorter after controlling for demographic variables; and, of note, had almost one hour less total night-time sleep. Conversely: they were significantly more likely to have more than one day-time nap (OR 7.75) and spent on average 17 min longer asleep during the day. However, additional day-time sleep did not compensate for shorter night sleep and overall children with DS experienced 37 min less sleep over 24 h than their TD peers.

3.7. Contribution of snoring to sleep patterns

Snoring status contributed significantly to the number and duration of night wakings, but not to any other night-time sleep variables (Tables 6 and 7, model 2). Children who snored were significantly more likely to wake more than once at night (OR 3.76) and spent half an hour longer awake at night than non-snorers. However, snoring did not fully explain group differences. Children with DS were still significantly more likely to have more than one night waking (OR 1.84) and spend longer awake at night than TD peers after controlling for snoring. It is noteworthy that, snoring history did not contribute significantly to longest uninterrupted sleep period at night, total night-time or total 24-hour sleep duration. While children who snored were significantly more likely to take more than one day-time nap (OR 5.05), their total time spent napping in the day was not significantly different to non-snoring children. Furthermore, children with DS were still significantly more likely to take a day nap (OR 5.07) after controlling for snoring status.

Table 3
Bedtime routines and settling practices.

	Number (%)		MODEL 1 group, age, gender, educational level	
	DS (n = 104)	TD (n = 489)	DS/TD OR (95%CI)	P[a]
Activities most nights in the hour before bedtime				
Bath	73 (70%)	416 (85%)	0.43 (0.26, 0.71)	0.002
Massage	12 (12%)	36 (7%)	2.32 (1.09, 4.90)	0.035
Reading books	49 (47%)	293 (60%)	0.46 (0.28, 0.74)	0.001
Being rocked	30 (29%)	47 (10%)	4.61 (2.60, 8.18)	0.000
Watching TV	30 (29%)	112 (23%)	0.99 (0.59, 1.66)	0.969
Dinner/snack	18 (17%)	42 (9%)	1.74 (0.92, 3.28)	0.096
Bottle/drink/nursing	79 (76%)	423 (86%)	0.65 (0.37, 1.13)	0.136
Running around	11 (11%)	67 (14%)	0.63 (0.31, 1.28)	0.188
Prayers	14 (14%)	17 (4%)	4.14 (1.89, 9.09)	0.001
Singing songs	30 (29%)	83 (17%)	2.08 (1.25, 3.46)	0.006
Listening to music	18 (17%)	61 (13%)	1.82 (0.99, 3.34)	0.061
How child falls asleep most of the time				
Bottle fed	29 (28%)	49 (10%)	4.75 (2.62, 8.64)	0.000
Breast fed	9 (9%)	49 (10%)	1.38 (0.62, 3.09)	0.442
Being rocked	19 (18%)	24 (5%)	5.87 (2.79, 12.36)	0.000
Being held	30 (29%)	38 (9%)	5.92 (3.26, 10.76)	0.000
Watching TV	6 (6%)	13 (3%)	1.40 (0.48, 4.04)	0.546
In own cot/bed alone	58 (56%)	368 (75%)	0.44 (0.27, 0.69)	0.001
In own cot/bed with parent	20 (19%)	51 (10%)	2.04 (1.11, 3.73)	0.026
In parents' bed with parent present	16 (15%)	19 (4%)	3.60 (1.70, 7.62)	0.001
Parent present at settling	44/79 (56%)	103/414 (25%)	4.40 (2.55,7.61)	0.000

[a] Likelihood ratio test for removal of DS/TD term from model 1.

Table 4
Sleep onset latency, consistent routine and bedtime difficulty.

		Number (%)		MODEL 1 group, age, gender, educational level		MODEL 1 + parent present group, parent present, age, gender, educational level	
		DS (n = 104)	TD (n = 489)	OR (95%CI)	P[a]	OR (95%CI)	P[a]
How long does it take for child to fall asleep?	<5 min	5 (5%)	47 (10%)				
	5–15 min	48 (47%)	292 (60%)				
	16–30 min	30 (29%)	121 (25%)				
	31–60 min	13 (13%)	25 (5%)				
	>1 h	6 (6%)	4 (1%)				
	>15 min	49/102 (48%)	150/489 (31%)				
	DS/TD OR			1.82 (1.15, 2.88)	0.011	1.42 (0.82, 2.47)	0.215
	parent present at bedtime/absent OR			(n = 591)		0.30 (0.19, 0.48) (n = 491)	0.000
Same bedtime routine?	never	4 (4%)	5 (1%)				
	1-2 nights/week	6 (6%)	6 (1%)				
	3-4 nights/week	14 (14%)	48 (10%)				
	5-6 nights/week	31 (30%)	178 (36%)				
	every night	49 (47%)	252 (52%)				
	DS/TD OR			0.82 (0.53, 1.28)	0.388	0.96 (0.56, 1.63)	0.865
	parent present at bedtime/absent OR			(n = 593)		2.31 (1.50, 3.56) (n = 493)	0.000
How difficult is bedtime?	very easy	45 (44%)	184 (38%)				
	somewhat easy	25 (24%)	162 (33%)				
	neither easy or difficult	19 (18%)	96 (20%)				
	somewhat difficult	13 (13%)	39 (8%)				
	very difficult	2 (2%)	8 (2%)				
	difficult[b]	15 (14%)	47 (10%)				
	DS/TD OR			1.40 (0.73, 2.71)	0.325	1.14 (0.52, 2.51)	0.747
	parent present at bedtime/absent OR			(n = 593)		0.19 (0.10, 0.39) (n = 493)	0.000

[a] Likelihood ratio test for removal of term from the specified model.
[b] Difficult = 'somewhat difficult' or 'very difficult'.

3.8. Contribution of parental presence at settling to sleep patterns

Parental presence at settling contributed significantly to all night-time sleep variables (Tables 6 and 7, model 3). On average children whose parents were physically present went to bed almost an hour later, were less likely to take longer than 15 min to fall asleep, were more likely to wake more than once (OR 2.70), and spent 11 min longer awake at night; yet, notably, their uninterrupted longest sleep period was over 2 h shorter. While day-time napping was not significantly influenced by parental presence at bedtime, night sleep and in turn total 24-hour sleep were around an hour shorter.

3.9. Residual differences in sleep patterns after controlling for snoring and parental presence

After controlling for both snoring status and parental presence at bedtime only four sleep outcomes remained significantly different between the DS and TD groups (Tables 6 and 7, model 3). In children with DS bedtime was 22 min later, and total night-time sleep was 38 min shorter. However, this was partly compensated for by more day-time naps and an average of 21 min longer spent asleep during the day. The net result of this shift of sleep from night to day was that 24-hour total sleep time was on average only 18 min shorter in DS children, a difference that did not reach statistical significance.

Table 5
Parental activities if child wakes during the night.

Circumstance	Number (%)		MODEL 1 group, age, gender, educational level	
	DS (n = 104)	TD (n = 489)	DS/TD OR (95%CI)	P[a]
Pick up and hold/rock back to sleep	42 (40%)	57 (12%)	6.41 (3.77, 10.89)	0.000
Pick up and put back whilst awake	22 (21%)	112 (23%)	0.99 (0.58, 1.71)	0.979
Rub/pat but do not pick up	35 (34%)	146 (30%)	1.23 (0.77, 1.97)	0.387
Bottle feed back to sleep	22 (21%)	61 (13%)	2.21 (1.23, 3.98)	0.010
Breast feed back to sleep	9 (9%)	52 (11%)	1.36 (0.61, 3.05)	0.467
Give a dummy	16 (15%)	46 (9%)	2.16 (1.12, 4.16)	0.027
Verbally comfort but do not pick up	25 (24%)	114 (23%)	0.91 (0.54, 1.53)	0.725
Bring into bed with parent	26 (25%)	66 (14%)	2.31 (1.34, 3.98)	0.003
Let child cry	12 (12%)	71 (15%)	0.79 (0.40, 1.55)	0.481
Give a few minutes to settle back to sleep	47 (45%)	243 (50%)	0.89 (0.57, 1.38)	0.595
Play with child	3 (3%)	3 (1%)	5.06 (0.94, 27.14)	0.070
Watch TV/DVD	3 (3%)	1 (0%)	13.54 (1.27, 144.66)	0.020
Sing to child	12 (12%)	28 (6%)	2.52 (1.19, 5.31)	0.021
Parent present until child falls back asleep	77 (74%)	273 (56%)	2.45 (1.49, 4.00)	0.000[1]

[a] Likelihood ratio test for removal of DS/TD term from model.

Table 6
Sleep patterns.

Outcome	Comparison	Means Unadjusted (adjusted from model 1)		MODEL 1 group, age, gender, educational level		MODEL 2 group, snoring, age, gender, educational level		MODEL 3 group, snoring, parent present, age, gender, educational level	
		DS	TD	Difference (95%CI)	Pª	Difference (95%CI)	Pª	Difference (95%CI)	Pª
Bedtime (minutes)	DS − TD	8h 9m (8h 10m)	7h 37m (7h 39m)s	31m (20, 43)	0.000	31m (18, 43)	0.000	22m (8, 35)	0.001
	snoring − not					3m (−17, 22)	0.789	0m (−20, 20)	0.973
	parent present at bedtime −absent							51m (42, 61)	0.000
				(n = 592)		(n = 592)		(n = 492)	
Duration of wakings (minutes)	DS − TD	29m (28)	11m (10)	17m (10, 24)	0.000	9m (2, 17)	0.014	8m (−1, 17)	0.094
	snoring − not					32m (20, 44)	0.000	39m (26, 52)	0.000
	parent present at bedtime − absent							11m (5, 18)	0.001
				(n = 590)		(n = 590)		(n = 491)	
Longest sleep (minutes)	DS − TD	7h 49m (7h 57m)	9h 19m (9h 22m)	−85m (−125, −45)	0.000	−71m (−115, −28)	0.001	−43m (−91, 5)	0.081
	snoring − not					−55m (−123, 12)	0.109	−50m (−122, 22)	0.175
	parent present at bedtime −absent							−140m (−175, −105)	0.000
				(n = 586)		(n = 586)		(n = 488)	
Night-time sleep (minutes)	DS − TD	9h 54m (9h 55m)	10h 50m (10h48m)	−53m (−72, −35)	0.000	−49m (−69, −29)	0.000	−38m (−61, −15)	0.001
	snoring − not					−16m (−47, 15)	0.311	−19m (−53, 15)	0.271
	parent present at bedtime − absent							−55m (−72, −38)	0.000
				(n = 591)		(n = 591)		(n = 491)	
Daytime sleep (minutes)	DS − TD	2h 2m (2h 5m)	1h 57m (1h 49m)	17m (5, 28)	0.004	14m (1, 26)	0.030	21m (7, 35)	0.003
	snoring − not					12m (−7, 31)	0.210	11m (−10, 32)	0.322
	parent present at bedtime − absent							−9m (−20, 1)	0.080
				(n = 592)		(n = 591)		(n = 492)	
Total sleep time (minutes)	DS − TD	11h 56m (12h 0m)	12h 47m (12h 37m)	−37m (−59, −15)	0.001	−36m (−60, −12)	0.003	−18m (−44, 9)	0.198
	snoring − not					−3m (−40, 33)	0.851	−8m (−48, 32)	0.703
	parent present at bedtime − absent							−64m (−84, −45)	0.000
				(n = 591)		(n = 591)		(n = 491)	

ª F test for removal of term from the specified model.

Table 7
Number of wakings during the night, naps during the day, and overall sleeping assessment.

		Number (%)		MODEL 1 group, age, gender, educational level		MODEL 2 group, snoring, age, gender, educational level		MODEL 3 group, snoring, parent present, age, gender, educational level	
		DS (n = 104)	TD (n = 489)	OR (95%CI)	P[a]	OR (95%CI)	P[a]	OR (95%CI)	P[a]
Number of night wakings	0	38 (38%)	197 (40%)						
	1	24 (24%)	190 (39%)						
	2	18 (18%)	73 (15%)						
	3	10 (10%)	19 (4%)						
	≥4	9 (2%)	10 (2%)						
	>1	37/99 (36%)	102/489 (21%)						
	DS/TD OR			2.48 (1.51, 4.07)	0.000	1.84 (1.07, 3.17)	0.031	1.25 (0.64, 2.44)	0.511
	snoring/not OR			(n = 588)		3.76 (1.66, 8.54)	0.002	3.54 (1.37, 9.16)	0.010
	parent present at bedtime/absent OR					(n = 588)		2.70 (1.65, 4.41)	0.000
								(n = 489)	
Number of naps during the day	0	3 (3%)	40 (8%)						
	1	45 (44%)	218 (45%)						
	2	34 (33%)	189 (39%)						
	3	12 (12%)	35 (7%)						
	≥4	9 (9%)	7 (1%)						
	>1	55/103 (53%)	231/489 (47%)						
	DS/TD OR			7.75 (3.48, 17.26)	0.000	5.07 (2.14, 12.01)	0.000	5.81 (2.18, 15.50)	0.000
	snoring/not OR			(n = 592)		5.05 (1.55, 16.39)	0.005	3.71 (1.07, 12.94)	0.037
	parent present at bedtime/absent OR					(n = 592)		1.57 (0.83, 3.00)	0.168
								(n = 492)	
How well does your child usually sleep at night?	very well	38 (37%)	275 (56%)						
	well	22 (21%)	116 (24%)						
	fairly well	25 (24%)	78 (16%)						
	fairly poorly	11 (11%)	15 (3%)						
	poorly	5 (5%)	4 (1%)						
	very poorly	3 (3%)	1 (0%)						
	poorly[b]	19 (18%)	20 (4%)						
	DS/TD OR			4.63 (2.25, 9.53)	0.000	2.89 (1.26, 6.65)	0.017	2.40 (0.85, 6.74)	0.107
	snoring/not OR			(n = 593)		5.14 (1.88, 14.07)	0.002	6.54 (2.05, 20.86)	0.002
	parent present at bedtime/absent OR					(n = 592)		2.00 (0.82, 4.87)	0.131
								(n = 492)	
Do you consider your child's sleep to be a problem?	a very serious problem	11 (11%)	4 (1%)						
	a small problem	35 (34%)	90 (18%)						
	not a problem at all	57 (55%)	395 (81%)						
	a problem[c]	46/103 (45%)	94/489 (19%)						
	DS/TD OR			3.04 (1.90, 4.87)	0.000	2.19 (1.30, 3.69)	0.004	1.56 (0.82, 2.94)	0.178
	snoring/not OR			(n = 592)		3.78 (1.71, 8.36)	0.001	3.91 (1.55, 9.83)	0.004
	parent present at bedtime/absent OR					(n = 592)		3.34 (2.02, 5.51)	0.000
								(n = 492)	

[a] Likelihood ratio test for removal of term from the specified model.
[b] Poorly = fairly poorly, poorly and very poorly.
[c] A problem = a very serious problem or a small problem.

3.10. Parent reported sleep problems

Parents of children with DS were more likely to report that their child had a sleep problem (45% v 19%) and this remained significant after controlling for both demographic variables (Table 7, model 1) and for snoring status (OR 2.19) (Table 7, model 2). However, after controlling for parental presence at bedtime (Table 7, model 3) differences between the groups were no longer significant suggesting that parental presence at bedtime significantly explained parental perception of sleep problems across the sample.

4. Discussion

To the best of our knowledge, this is the first study to explore possible reasons for sleep differences between infants and toddlers with Down syndrome (DS) and typically developing (TD) children. In support of previous research, we found clinically important differences in sleep in DS children. Notably, on average they had almost an hour less sleep at night than TD peers and, despite slightly more sleep in the day, were still sleeping, on average, 37 min less over 24-hours. However, two fundamental differences in the groups were observed which could have partly, or wholly, explained these differences.

First, in support of the established literature [5], we confirmed a higher prevalence of habitual snoring in the DS children (19% vs 2%). It is well established that children with DS are vulnerable to sleep disordered breathing due to their unique craniofacial anatomy and low muscle tone. While sleep disordered breathing may have less impact on sleep quality in TD children than previously assumed [27], it remains an important potential cause of poor sleep in children with DS [4].

Second, we observed striking differences in sleep ecology between the groups. While regular bedtime routines were reported for most children, a pattern of effortful parental involvement was more likely in children with DS, with activities such as rocking and massaging more often included in the bedtime routine. It is noteworthy that, parents of children with DS were significantly more likely to stay with their child until they fell asleep at bedtime (OR 4.40) and after night wakings (OR 2.45) using techniques such as rocking or bottle feeding to settle their child. Similar findings were reported by Bassell et al., in 32 toddlers with DS [21], a third of whom needed a parent in the room to fall asleep. Of note, the study included children up to 13 years and this 'need' for parental presence did not diminish with age.

The contribution of these two key factors to sleep differences between the groups are an important consideration. While snoring only increased the likelihood of night waking, parental presence at bedtime was associated with later bedtimes, more frequent and longer night wakings, shorter periods of uninterrupted sleep, no compensatory increase in day naps, and an average of one hour less 24-hour sleep overall. The cross-sectional study design means that the direction of this association cannot be assumed. Parental presence may have driven differences in children's sleep or the converse may be true. Nonetheless, an evidence based, theoretical framework supports the possibility that parental presence at bedtime may influence children's sleep behaviors. Parental behavior at bedtime has been consistently demonstrated to predict settling difficulties and night waking in young children [28]. Based on both learning theory and operant conditioning, children 'learn' to self-soothe to sleep within a familiar environment. Settling strategies such as rocking a child to sleep are highly rewarding for the child and, while they may be effective, they limit the child's ability to learn self-soothing skills [29], leading to unintended consequences of night-waking. Parental behavior is driven by many factors including cultural norms, beliefs about the child, the child's

health and temperament, environmental factors, and parental mental health. Children with DS are at increased risk neuropsychiatric co-morbidities such as autism spectrum disorder (ASD) and attention deficit hyperactivity disorder (ADHD) [30]. Furthermore, while these co-morbidities are rarely diagnosed in infants and toddlers, behavioral features are evident before three years of age [31]. Such behaviors will prompt adaptive parenting styles and could explain the effortful approaches to settling seen more frequently in the parents of children with DS. Furthermore, these co-morbidities are independently associated with sleep disorders, (eg, sleep-related movement disorders) which could have contributed to the sleep pattern differences. Future research should include measures of ASD and ADHD features to examine the extent to which these comorbidities affect both parenting style and sleep patterns. Parents of children with developmental disorders may have low expectations of their child's sleep [32]. Normalisation of sleep problems may explain the delay, or failure, to seek specialist help [33]. This may be compounded by lack of skills and knowledge about children's sleep in health professionals [34], allowing unhelpful practices to become embedded. Furthermore, studies of children with long-term conditions report parental 'chronic sorrow' [35] which may prompt a more permissive parenting style [36]. Fear of night-time epilepsy, or apnea [37], and concerns about the child's physical health may prompt vigilant behavior. Indeed, in this study children with DS were more likely to share a parent's bedroom and be taken into their parents' bed after night waking. In a study of school-aged children with DS, 48% were reported to regularly move into their parents' bed during the night [18], suggesting that these behaviors may persist through childhood. Co-sleeping practices in parents of children with complex needs result in a shared burden of poor sleep with parents also suffering from sleep loss [38]. Chronic sleep deprivation adds to parenting stress and risk of maternal depression [39], resulting in a vicious cycle which limits parents' capacity to address the primary problem.

Finally, it should be noted that there were residual differences in sleep patterns between the groups not explained by snoring or parental presence at bedtime. Specifically, children with DS settled to sleep later, had 38 min less sleep at night but 21 min more sleep in the day. Moreover, the differences in 24-hour total sleep time were eliminated by this shift of sleep from night to day. These differences in circadian sleep distribution are intriguing. A previous study of circadian phase in 66 children with DS aged 5–67 months showed a phase-advanced pattern in 35% of the sample [22] rather than the tendency towards phase delay reported here. It is likely that non-biological mechanisms explain our findings. First, a higher prevalence of bedtime resistance has been previously described in children with DS [18], and, independent of parental presence at bedtime, could lead to later sleep onset times. Social factors could also contribute. Mothers of children with disabilities are less likely to be in paid employment [40] and may continue to promote longer day naps than mothers who return to work. Reduced sleep opportunity at night due to late bedtimes, compounded by worse sleep quality [22], could explain the high prevalence of excessive daytime sleepiness frequently reported in this population [18,21]. While extended nap times offer an opportunity for compensatory sleep in infants and toddlers, by school-age this is no longer possible, underlining the need for early intervention to optimise night-time sleep.

4.1. Limitations

The two groups were surveyed at different times, children with DS were studied 6–9 years after the TD control group. Although historic data suggests a reduction in total sleep time over the past

century of around one hour [41], recent data from Australian school-aged children indicates that sleep duration remained stable between 1985 and 2012 [42]. It is unlikely that any social change in the UK in the period 2006 to 2015 would have influenced parenting practices for pre-schoolers to the extent reported here.

Selection bias is possible in both samples as parents with concerns about their child's sleep may have been more likely to participate in research with a sleep focus. Due to the large sample size of TD children, assessment of sleep was limited to parental questionnaire with inherent risks of response bias. Use of objective measures such as actigraphy in future studies could usefully supplement parental report.

Finally, snoring 'always or almost always' was used as the best proxy measure for sleep disordered breathing. Snoring is the most reliable single symptom of sleep disordered breathing but is a poor predictor of apnea severity [43,44]. Objective measurement of sleep disordered breathing was impractical in the large sample size available here. The impact of treating both sleep apnea and behavioral insomnia on sleep quality in this population should be evaluated in future trials.

4.2. Implications for practice

Children in this study who settled to sleep with a parent present had, on average, one hour less sleep per 24 h. Restricted sleep in the early years has potential implications for future neurodevelopment [16] and children with DS may be particularly vulnerable due to their limited cognitive reserve [45]. Indeed, recent data in pre-school children with DS support an association between sleep problems and language development and between obstructive sleep apnea and executive function deficits [46,47].

While longitudinal sleep data in children with DS are lacking, cross-sectional studies suggest persistence of sleep problems through childhood and adolescence [21]. Our data suggest that DS treatment programs should actively screen for sleep problems. Where problems are identified, specialist evaluation should look beyond sleep disordered breathing. Notably, our data suggest that these children may be particularly vulnerable to behavioral insomnia related to parental presence at bedtime and an inability to return to sleep after natural awakenings, and these problems may have a particularly erosive impact on their sleep. Prevention is better than cure and early sleep education for parents of infants with Down syndrome should be developed to encourage healthy sleep habits for life.

Conflict of interest

Dr. Mindell reports grants and personal fees from Johnson & Johnson, during the conduct of the study.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2019.05.005.

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