NEW RESEARCH

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Body Position and Obstructive Sleep Apnea in Children with Down Syndrome

Egambaram Senthilvel, M.D.; Jyoti Krishna, M.D.

Neurological Institute, Sleep Disorder Center, Cleveland Clinic Foundation, Cleveland, OH

Introduction: Children with Down syndrome (DS) commonly have obstructive sleep apnea syndrome (OSAS) and may assume a unique sleeping position not systematically described previously. We describe this sleep position in DS and explore its relationship with OSAS in comparison to control children (CC).
Methods: Overnight video-polysomnograms (PSG) of consecutive children with DS (age 2-18 y), referred to our center between April 2008 and October 2009, were retrospectively analyzed by a single scorer (ES). CC group comprised age and gender matched, non-syndromic, neurologically intact children referred to us for suspected OSAS over the same period.
Results: Each group had 17 subjects matched for age (median [IQR]; 6 [4-8]) and gender (65% female). DS group had higher BMI (median [IQR]; 18.8 [17.4-21.0]) than CC (17 [14.7 -18.8]; p = 0.03). There were however no significant differences (median [IQR]) between DS and CC with respect to sleep time in minutes (460 [425-499] vs 424[410-483]), sleep efficiency (%) (90.9 [87.4-92.4] vs 88.6 [79.9-93.1]), REM time (%) (17.1 [14.2-22.1] vs 19.2 [14.9-22.1]), supine time (%) (40.7 [24.8-56.0] vs 15.8 [0.40-44.5], p 0.06), mean oxygen saturation (%) (95 [94-96] vs 96 [95-97]), oxygen saturation nadir (89 [86-91] vs 89[94-92]), or total apnea-hypopnea index (4.3 [3-7.8] vs 5.1[1.9-9.6]). Despite these similarities between the groups, 9 (53%) DS children slept seated bent forward with head resting on bed for at least part of the total sleep time (%) (7.8 ± 10.9, range 0.8-35.7).This was absent in the CC group (p = 0.06).
Conclusion: Some DS children assume a peculiar body position, sitting cross-legged flopped-forward with head resting on bed while asleep. This is absent in age- and gender-matched controls showing otherwise similar PSG characteristics. The reason for this posture is unclear from this study. However, this novel report of a unique sleeping position would provide us with a basis to conduct a prospective study involving a larger population to ascertain the contribution of this position for OSAS protection or to determine if it may be *forme fruste* parasomnia.
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SCIENTIFIC INVESTIGATIONS

Obstructive sleep apnea syndrome (OSAS) is a relatively common sleep disorder in the general pediatric population with estimated prevalence of 1% to 3%,[1-3] and it occurs in 45% to 79% of patients with Down syndrome (DS).[4-9] Children with DS are prone to develop OSAS due to a combination of multiple anatomical and physiological factors.[10] The relationship between OSAS and body position during sleep has been studied in adults where it is generally known that supine position increases propensity to apnea events[11,12] compared to off-supine positions.[13-15] However in children, significant disparities exist from the several studies conducted thus far on body position and obstructive events during sleep.[16-20]

It has been our clinical impression that parents of children with DS frequently report a unique body position (sitting cross-legged and leaning forward) during sleep, perhaps as a mechanism for airway protection. Further, while obstructive sleep apnea in children with DS has been described in several studies,[4-8] to the best of our knowledge, the effect of body position on obstructive sleep apnea in children with DS has not been described. Our objectives in the present study were: (1) To evaluate the prevalence of this unique sleeping position in children with DS compared to matched controls (CC); and (2) To evaluate if this unique sleeping position has any protective role in OSAS.

BRIEF SUMMARY
Current Knowledge/Study Rationale: A peculiar sitting cross-legged with torso flopped forward sleeping position is a common concern of parents of children with Down syndrome. However, to the best of our knowledge this has never been previously described in the literature.
Study Impact: We systematically describe this phenomenon for the first time in relation to control subjects utilizing polysomnography. We speculate if this may be *forme fruste* parasomnia or perhaps a means to protect an airway prone to obstructive sleep apnea.

METHODS

Study Groups

The study was performed at Cleveland Clinic Sleep Disorders Center after prior approval by the institutional review board. We retrospectively reviewed consecutive overnight PSGs of children with DS performed between April 2008 and October 2009. We included all DS children between 2 and 18 years of age. The lower age limit of 2 years was used as cut-off to allow for maturation of motor skills in DS, so that there was a reasonable possibility of upright posturing in sleep. Consecutively presenting age- and sex-matched children who had no apparent genetic or clinically significant morphologi-

cal abnormalities and underwent PSG for suspected sleep apnea over the same period of time, were identified as controls (CC). Children who did not have video data in our archives (to visually confirm body position) were excluded, as were those whose sleep efficiency was < 50%. Data retrieved from the sleep studies and clinical electronic medical records included age, gender, height, weight, BMI, ethnicity, tonsillar size (graded from 1 to 4), history of adenotonsillectomy, morphological deformities, and comorbid medical conditions. Body position and PSG data of the children with DS were analyzed by a single scorer (ES) and compared with the control group.

Polysomnogram

Children were studied in a dark, quiet room in the company of a parent in a single accredited sleep laboratory using Nihon Kohden America, Inc., PSG systems. Bed sharing with parent was not allowed. Standard leads were used to record electroencephalogram, electrooculogram, and electromyogram (submental and anterior tibialis). Respiratory parameters were measured by thoracic and abdominal inductance plethysmography belts; oronasal air flow was measured using a nasal pressure transducer cannula and thermistor. Pulse oximeter, end-tidal and transcutaneous carbon dioxide sensors, EKG, and snore microphone were utilized. Overnight digital video was recorded using infrared camera for the entire portion of the sleep study, which was also fully attended by a sleep technician.[21]

Sleep studies were scored by registered sleep technologists and reviewed by a sleep physician (ES) using standard pediatric criteria. The 2-breath rule was used for obstructive event scoring.[21] Obstructive apneas were defined as reduction of airflow of > 90% from baseline amplitude with continuing effort to breathe. Hypopneas were defined as a reduction in airflow ≥ 50% from the baseline associated with desaturation ≥ 3%, arousal, or awakening. The apnea hypopnea index (AHI) was defined as the number of apneas or hypopneas occurring per hour of sleep time.

Body Position

Body position was determined by reviewing the real-time digital video of the PSG in 30-sec epochs by a single scorer (ES). Further, where gross body movement was apparent on PSG channels, the video was played to mark the change of body position accurately. Body position was categorized as being supine, right side, left side, prone, upright, and leaning forward, based on the position of the torso. Percentage of total sleep time (TST) time spent in each position as well as AHI in each position was calculated.

Data Analysis

Continuous variables were presented as median (IQR), and categorical variables as fractions (%). Differences between Down syndrome and control groups were tested with the Wilcoxon rank sum test for continuous variables and the Fisher exact test for categorical variables. We also compared patient characteristics between Down syndrome patients who leaned forward and who did not using similar analysis. A p-value < 0.05 was considered statistically significant. S-Plus 7.0 (Insightful, WA) was used for all statistical analyses.

Table 1—Comparison of Down syndrome (DS) and control (CC) characteristics

Variables	DS (n = 17)	CC (n = 17)	p value
Age median (IQR)	6 (4-8)	6 (4-8)	0.9
Gender (%)			
Male	35	35	1
Female	65	65	
Race (%)			
Caucasian	88	65	0.1
African American	6	35	
Hispanic	6	0	
BMI median (IQR)	18.8 (17.4-21.0)	17.0 (14.7-18.8)	0.03*
Tonsil Size (%)			
Grade 0 (absence secondary to tonsillectomy)	41	24	0.7
Grade 1	0	0	
Grade 2	12	12	
Grade 3	41	59	
Grade 4	6	5	
Comorbidity (%)			
History of congenital heart disease	82	12	< 0.0001*
History of hypothyroidism	41	0	0.003*

*p value significant. Wilcoxon rank sum test for continuous variables and the Fisher exact test for categorical variables.

RESULTS

A total of 25 children with DS were identified. However, 7 children were excluded due to lack of video for review, and one child younger than 2 years of age was excluded per study protocol. Therefore, the study population included 17 children with DS; 17 consecutive controls meeting inclusion criteria were selected as described. Unless specified, data is presented as median (interquartile range, [IQR]) (**Table 1**). The age was 6 years [4-8], and females (65%) and Caucasians were predominant in both groups. Children with DS group had significantly higher BMI than control group (18.8 [17.4-21.0] vs 17.0 [14.7-18.8)] p = 0.03). History of prior tonsillectomy was found in 41% of DS compared to 24% of CC. Not surprisingly, a history of congenital heart disease (of any type) was present in 82% of DS against 12% of CC (p < 0.0001). Similarly, history of hypothyroidism was present in 41% of DS compared to 0% of CC (p = 0.003). See **Table 1** for detailed information.

There were no significant statistical differences between DS and CC with respect to sleep characteristics and presence of sleep disordered breathing (**Table 2**). Despite these similarities between the DS and CC groups, 9 (53%) of DS children slept seated and leaning forward with their head resting on the bed (**Figure 1**) for at least part of the total sleep time (TST) (7.8% ± 10.9%, range 0.8-35.7). However, this was never seen in the CC group (p = 0.06). DS also spent more time in supine (median [IQR] 40.7% TST [24.8-56.0]) compared to CC (15.8% [0.40-

Table 2—Sleep characteristics of DS and CC

Variables	DS (n = 17)	CC (n = 17)	p
Total sleep time (min)	460 (425-499)	424 (410-483)	0.4
Sleep efficiency %	90.9 (87.4-92.4)	88.6 (79.9-93.1)	0.6
REM time %	17.1 (14.2-22.1)	19.2 (14.9-22.1)	0.9
Supine time %	40.7 (24.8-56.0)	15.8 (0.40-44.5)	0.06*
Right side %	18.1 (0.95-30.8)	18.6 (15.1-42.3)	0.4
Left side %	5.9 (0.1-18.0)	29.7 (5.4-42.2)	0.04
Prone %	6.1 (0-23.9)	11.7 (0-27.9)	0.6
Upright %	0.01 (0-0.1)	0 (0-0)	0.3
Leaning forward (n, %)	9 (53)	0	0.06*
Mean O_2 saturation %	95 (94-96)	96 (95-97)	0.2
O_2 nadir %	89 (86-91)	89 (84-92)	0.9
Total AHI	4.3 (3.0-7.8)	5.1 (1.9-9.6)	0.9
Arousal index	16.3 (13.3-19.9)	14.7 (12.6-20.7)	0.8
No of positions changed	22 (18-33)	15 (13-27)	0.2

*Statistical significance approaching 0.05. Continuous variables expressed as median (IQR)

Figure 1—Polysomnographic video frame of a patient showing peculiar sitting-flopped-forward body position with head resting on bed while asleep

44.5]; p = 0.06). The overall AHI did not differ significantly between DS and CC. The AHI was also similar in both groups in the various body positions.

The characteristics of children with DS who adopted the leaning forward (DSLF) position at any time during the PSG were generally similar to those DS who did not lean forward (DSNLF) (**Tables 3**, **4**), with the exception that significantly more DSLF children than DSNLF had some form of congenital heart disease by history (100% vs 63%; p = 0.04). A history of tonsillectomy (67% vs 12%) and hypothyroidism (56% vs 25%) appeared to be more common in DSLF but did not reach statistical significance. Further, PSG characteristics of children with DSLF vs DSNLF were very similar in regard to TST, sleep efficiency, REM time, mean oxygen saturation, total AHI, and number of body position changes. However, DSLF spent less time in supine position compared to DSNLF (24.8 [16.9-37.0] vs 50.5 [43.3-64.9]; p = 0.01). AHI was lowest in prone and leaning forward in children with DS. AHI in different body positions did not reach statistical significance when compared between DSLF and DSNLF and within DSLF (**Table 5**). Further, the time spent leaning forward and severity of AHI were not significantly correlated (Spearman r = −0.20, p = 0.5).

Of the 9 DSLF, 4 attained this posture in stage 3, 3 in stage 2, 1 in stage 1, and 1 in REM sleep. The assumption of this posture was not associated with sleep talking or confusional behaviors.

DISCUSSION

In the current study, we found that BMI was higher in children with DS when compared to CC. However, the overall AHI between both groups were similar. This finding is consistent with those of de Miguel-Díez et al.[5] and Fitzerald et al.[8] who reported that increased severity of sleep disordered breathing in children with DS was not significantly associated with high BMI. Our study shows that congenital heart disease and hypothyroidism were common in children with DS, as previously described in the literature.[22,23]

The main finding of this study is that some DS assumed a unique sitting-flopped-forward body position with head resting on bed while asleep. This was absent in controls showing otherwise similar PSG characteristics. This body position has not been described previously in the literature.[4-9] In comparing the sleep characteristics of DS with CC, none of the sleep variables were statistically significant. However, children with DS overall spent more time in supine position than CC; the difference approached statistical significance (p = 0.06). Similarly, leaning forward position was not present at all in CC compared to children with DS. This finding also approached, but did not achieve statistical significance, likely because of small sample size. However, we also noted that several parents of DS children specifically mentioned this position during the initial presentation to the sleep clinic, although we did not systematically collect data on these parental reports. The reason for this position is unclear from our study, and it can only be conjectured that this may be a protective mechanism for airway patency or *forme fruste* parasomnia. It was noted to occur most often in stages N3 and N2.

There were no differences found between DS and CC with respect to TST, sleep efficiency, REM time, mean oxygen saturations, saturation nadir, or total AHI. But DSNLF spent more time in supine position than DSLF. Only history of heart disease was more commonly present in DSLF compared to DSNLF. Again, because a limited number of patients in our study, we do not know the significance of this comorbid association with this unique position.

From this study, the reason for this posture appears to be not related to gender, BMI, or hypothyroidism between DS children with and without leaning forward position. Nor does it appear to be a marker for the presence or severity of OSA in Down syndrome patients. However, our novel finding of this unique sleeping position provides us with a basis in the future to conduct a prospective study involving a larger population of

Table 3—Comparison between DS patients who did and did not lean forward

Variables	Leaning forward DS (n = 9)	Non leaning forward DS (n = 8)	p value
Age, median (IQR)	5.0 (4.0-10.0)	7.0 (3.8-7.3)	0.9
Male, n (%)	4 (44)	2 (25)	0.4
BMI, median (IQR)	18.8 (17.7-19.2)	19.6 (1.70-22.9)	0.8
Tonsillectomy, n (%)	6 (67)	1 (12)	0.1
Heart disease, n (%)	9 (100)	5 (63)	0.04*
Hypothyroidism, n (%)	5 (56)	2 (25)	0.2

*p value significant. Continuous variables expressed as median (IQR).

Table 4—Sleep variables of DS who did and did not lean forward

Variables	Leaning forward DS (n = 9)	Non leaning forward DS (n = 8)	p value
Total sleep time (min)	425 (414-496)	472 (453-512)	0.2
Sleep efficiency %	91.5 (84.4-92.4)	90.8 (89.4-92.3)	0.8
REM time %	16.7 (15.5-22.1)	17.5 (14.1-22.5)	0.9
Supine time %	24.8 (16.9-37.0)	50.5 (43.3-64.9)	0.01*
Mean O_2 saturation %	94 (94-96)	95 (94-96.5)	0.5
Total AHI	3.9 (3.0-6.6)	4.5 (3.1-8.2)	0.6
No of position changes	22 (20-33)	19.5 (17.8-33.3)	0.7

*p value significant. Continuous variables expressed as median (IQR).

Table 5—AHI in different body positions in DS who did and did not lean forward

Variable	Leaning forward DS (n = 9)	Non leaning forward DS (n = 8)	p value
Total AHI	3.9 (3.0-6.6)	4.5 (3.1-8.2)	0.6
Supine AHI	5.8 (2.4-11.8)	5.6 (4.0-7.4)	0.9
Right AHI	2.3 (1.6-6.0)	2.4 (1.7-4.3)	0.9
Left AHI	2.5 (1.2-2.7)	1.4 (0.08-4.3)	0.8
Prone AHI	1.8 (1.1-2.9)	1.3 (0-7.5)	0.8
Upright AHI	0 (0-1.3)	0	1.0
LF AHI	1.8 (0-7.2)	NA	NA
Total off supine AHI	2.6 (1.7-3.9)	4.6 (3.0-6.5)	0.5

All variables expressed as median (IQR). NA = not available.

children with Down syndrome to further analyze the contribution of this position for OSAS protection, to examine if it may be *forme fruste* parasomnia, and further explore if this phenomenon is noted in children with other etiologies for delayed development or neurobehavioral disorder.

ABBREVIATIONS

DS, Down syndrome
OSAS, Obstructive sleep apnea syndrome
CC, Control children
PSG, Polysomnogram
IQR, Interquartile range
AHI, Apnea hypopnea index
TST, Total sleep time
DSLF, Down syndrome leaning forward
DSNLF, Down syndrome non leaning forward

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Address correspondence to: Egambaram Senthilvel, 2505 Ithaca Drive, Avon, OH 44011; E-mail: senthilvelegambaram@yahoo.com

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