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Neurobehavioral Outcomes in School-Aged Children with Primary Snoring

Abstract

Objective

We assess behavioral and attentional problems and neurocognitive functioning in school-aged children with primary snoring (PS).

Methods

Seventeen children with PS and 27 non-snoring peers aged 6–10 years took part in the study. All children underwent a polysomnography (PSG) at the Sleep Laboratory. Snoring was defined by parents and with PSG. Children with obstructive sleep apnea were excluded. The parents completed the Sleep Disturbance Scale for Children. Parents and teachers assessed behavioral and attentional problems with the Child Behavior Checklist and the Conners' Rating Scale-Revised. Neuropsychological assessment included the Wechsler Intelligence Scale for Children and the Developmental Neuropsychological Assessment (NEPSY test).

Results

The PS group had significantly more parent-reported internalizing, total, and attentional problems than the control group. Teachers did not report behavioral problems in the PS group. The PS and control group had equal IQ scores and similar core neurocognitive functions, except for one visuospatial subtest. The PS group had significantly more inspiratory flow limitation and increased diaphragmatic electromyography compared with the controls. Parents reported significantly more daytime sleepiness in the PS group. Daytime sleepiness and snoring time were consistently associated with more behavioral and attentional problems. Flow limitation and more oxygen saturation values under 90% were associated with attentional problems, higher oxygen desaturation index and lower mean oxygen saturation percentage with reduced language functions.

Conclusions

Snoring with an increase in respiratory effort without apneas and hypopneas and parent-reported daytime sleepiness may be linked to daytime symptoms. School-aged children with PS are at risk for behavioral and attentional problems, but not cognitive impairments.

Introduction

Sleep disordered breathing (SDB) is common in childhood and covers breathing abnormalities from primary snoring (PS) to obstructive sleep apnea syndrome (OSA) (Sinha & Guilleminault, 2010). The prevalence of OSA in school-aged children is reported to be between 1 and 4% (Kaditis et al., 2004; Lumeng & Chervin, 2008). The prevalence of habitual snoring is, however, higher and varies widely, ranging from 4 to approximately 20% (Brockmann et al., 2012a; Ferreira et al., 2000; Kaditis et al., 2004). The term primary snoring is used when habitual snoring is not associated with apneas, hypopneas, frequent arousals, or gas exchange abnormalities (Kaditis et al., 2016).

Behavioral and attentional problems are reported in school-aged children with PS (Beebe, Ris, Kramer, Long, & Amin, 2010; Bourke et al., 2011b; Miano et al., 2011; O'Brien et al., 2004). The most compelling evidence regards hyperactivity, inattention, and social problems (Biggs, Nixon, & Horne, 2014; Brockmann, Urschitz, Shlaud, & Poets, 2012b; Miano et al., 2011; O'Brien et al., 2004). A few studies have found that children with PS have internalizing problems, showing more anxious/depressed mood, somatic complaints, and withdrawn/depressed symptoms (Beebe et al., 2010; Bourke et al., 2011b; O'Brien et al., 2004). However, conflicting results also exist. Blunden, Lushington, Kennedy, Martin, and Dawson (2000) and O'Brien et al. (2004) found no group differences in social competency or problematic behavior between children with SDB and controls. Findings on behavioral outcomes have mainly been based on parent reports. Studies using a multi-informant approach have shown that parents of children with PS typically report a greater severity and range of problematic behavior than teachers (Ali, Pitson, & Stradling, 1993; Arman et al., 2005; Beebe et al., 2004; Kohler, Kennedy, Martin, & Lushington, 2013; O'Brien et al., 2011).

Despite growing evidence suggesting compromised cognitive functioning in children with PS, the results have been inconsistent. Indeed, some studies report impairments in verbal and non-verbal reasoning, memory, and executive function in children with PS compared with healthy controls (Blunden et al., 2000; Bourke et al., 2011ab; Hunter et al., 2016; Kennedy et al., 2004; Miano et al., 2011; Morsbach Honaker, Gozal, Bennett, Capdevila, & Spruyt, 2009; O'Brien et al., 2004), whereas others (Beebe et al., 2010; Calhoun et al., 2009; Jackman et al., 2012; Maski et al., 2017) do not confirm these findings. Further, even though lower cognitive function is reported in children with PS compared with controls, the scores are usually within normal range (Biggs et al., 2014; Miano et al., 2011). This questions the clinical significance of cognitive impairment related to PS.

In some studies, children with PS are reported to exhibit even poorer behavioral outcomes (Jackman, et al., 2012; Smith et al., 2016) and similar neurocognitive impairments than children with more severe SDB (Bourke et al., 2011a; Miano et al., 2011). This has led to suggestion that PS may represent a unique phenotype of SDB instead of being its mildest form (Biggs et al., 2014). The underlying mechanisms of neurobehavioral deficits in children with PS are not fully understood. Galland et al. (2015) have suggested that in addition to the apnea-hypopnea index (AHI), flow limitation and increased breathing effort may contribute to these deficits, but these are not usually detected. In OSA, AHI is reported to associate with lower intellectual function (Jackman et al., 2012) and other higher cognitive functions (Hunter et al., 2016), but the findings are not consistent (Bourke et al., 2011a). Indeed, it has been presented that snoring rather than AHI severity might predict the likelihood of cognitive problems in 4- to 10-year-old children (Smith, Gozal, Hunter, & Kheirandish-Gozal, 2017). Another significant factor possibly affecting neurobehavioral outcomes in PS is daytime sleepiness. Parents have consistently recognized on questionnaires that children with SDB are sleepier than controls, but when objectively testing sleep propensity, they rarely fall in the range considered to be pathologic in adults (see Beebe, 2006).

Despite the varying findings regarding exposure to SDB response reported in earlier studies, evidence suggests that snoring affects the daytime functioning of children with PS. The problem, however, is that PS definitions vary and interfere with reaching a comprehensive view. In many studies, the definition of snoring relies on parent-reported snoring only. As a result, some of the included children may also have OSA in addition to snoring (Arman et al., 2005; Brockmann et al., 2012a; O'Brien et al., 2011). In some studies, PSG was performed to exclude OSA, but snoring was not verified (Beebe et al., 2010; Biggs et al., 2011; Bourke et al., 2011ab; Jackman et al., 2012; Morsbach Honaker et al., 2009; Smith et al., 2016; Smith et al., 2017) or the amount of snoring was not quantified (Ali et al., 1993; Blunden et al., 2000; Calhoun et al., 2009; Frye et al., 2018; Kennedy et al., 2004; Miano et al., 2011; O'Brien et al., 2004). To the best of our knowledge, snoring time has been quantified in only one previous study that reported that memory consolidation does not differ between children with PS and healthy subjects (Maski et al., 2017).

The present study aims to clarify the impact of PS on neurobehavioral performance. This was achieved by first selecting children with habitual snoring based on parent reports, excluding children with OSA, and then quantifying the snoring time from the PSG signals. In addition, flow limitation and increased respiratory effort were observed. We assumed that this more detailed detection of breathing abnormalities would find more consistent associations with neurobehavioral functioning. Neuropsychological assessment was conducted with standardized tests and a multi-informant approach on behavioral ratings was used to obtain a more comprehensive view of a child's behavior.

The focus of this study was on school-aged children, and the aims were to assess both the behavioral and neurocognitive problems, and to investigate associations between sleepiness, snoring, and neurobehavioral implications. Children with PS were expected to have elevated scores on problem behavior, especially in attention and internalizing problems (Beebe et al., 2010; Bourke et al., 2011b; Brockmann et al., 2012b; Miano et al., 2011; O'Brien et al., 2004). Also, some discrepancies on parent and teacher behavioral ratings were anticipated (Arman et al., 2005; Kohler et al., 2013; O'Brien et al., 2011). In addition, it was hypothesized that children with PS perform worse than nonsnoring peers in neurocognitive functioning, showing a diffuse pattern with mild impairments, e.g., in verbal and non-verbal reasoning, memory, and executive function when compared with healthy peers (Blunden et al., 2000; Bourke et al., 2011ab; Hunter et al., 2016; Kennedy et al., 2004; Miano et al., 2011; Morsbach Honaker et al., 2009; O'Brien et al., 2004).

Methods

Participants and Procedure of the Study

The present study is a part of a larger study evaluating sleep in school-aged children. The seventeen largest primary schools out of a total of 32 primary schools located in the city of X, Finland, were randomly selected. Three primary schools for children with hearing impairment, motor skill disorder, specific language disorder, and mental disability were excluded. All the parents of children (aged 6–10 years) enrolling in first- or third-grade classes were given a questionnaire that asked for demographics and background data and included the Finnish version of the Sleep Disturbance Scale for Children (SDSC), Bruni et al., 1996). SDSC is an instrument for assessing sleep problems and snoring in school-aged children. The questionnaire was handed out during physical examination by the school nurse or in class by the teacher and parents filled out the form at home. Daytime sleepiness was assessed using the SDSC factor Disorders of Excessive Somnolence (including the items:

difficulty in waking up, tired when waking up, sleep paralysis, daytime sleepiness, sleep attacks during the day).

The sampling procedure is presented in Figure 1. Of the 831 families, 333 (40%) returned the sleep questionnaire. Four children had to be excluded because of missing snoring information. Thus, the analyses comprised 329 children (184 first-graders and 145 third-graders). Children were divided into two groups based on parent-reported snoring. Children with snoring (n = 33) had to snore at least three nights a week, whereas children without snoring (n = 259) were allowed to snore only 1 to 2 nights a month. Children who snored 1 to 2 nights a week were excluded. Thirty-five children without snoring and 27 children with snoring were willing to participate in the clinical study with PSG and neuropsychological assessment. The study was approved by the Ethical Committees of X University Hospital and the City of X, and the parents of the recruited children gave their written informed consent.

Polysomnographic Recordings and Analyses

In total, 62 children underwent a PSG in the Sleep Laboratory at X University Hospital with an Embla N7000 device (Embla®, Natus Medical Inc., USA) after one adaptation night. PSG comprised six EEG derivations (Fp1-A2, Fp2-A1, C3-A2, C4-A1, O1-A2 and O2-A1), two electro-oculography (EOG) channels, submental muscle tonus, electrocardiogram, airflow by nasal pressure transducer and thermistor, thoracoabdominal respiratory movements, electromyography (EMG) from the abdomen and diaphragm, oxygen saturation with pulse oximetry, transcutaneous carbon dioxide tension ($TcCO_2$), leg movements, body position, and piezo snoring sensor on the neck. The sleep staging was performed with the standard rules (Iber, Ancoli-Israel, Chesson, & Quan, 2007) by two independent clinical neurophysiologists. A senior neurophysiologist performed the consensus scoring. The level of agreement between the two independent scorers was 85.8% (Kappa = 0.76).

Arousals were scored according to the criteria of the American Sleep Disorders Association (Bonnet et al., 1992). The obstructive apnea-hypopnea index (OAHI) was calculated as the number of obstructive apneas and hypopneas per hour (Berry et al., 2012). Snoring was quantified from the nasal pressure signal and piezo sensor and ensured from the video signal. Snoring episodes lasting more than 3 consecutive breathing cycles were selected and the snoring time (percentage referred to total sleep time) was defined. In addition, periods with sustained inspiratory flow limitation (FL) were selected. In the scoring of sustained FL, all 2-minute epochs containing at least 90% of flattened (non-round) inspiratory flow shape in the nasal pressure signal were selected. The 2-minute epoch scoring was also used to select periods with increased EMG in the diaphragm electromyography signal.

For this study, the children were divided into two groups, a PS group and a control group, based on the respiratory variables. In addition to parent-reported snoring (snored at least three nights a week to every night), the inclusion criteria for the PS group were an OAHI < 1/h and PSG snoring time $\geq 1.5\%$. Ten children with snoring were excluded from the analyses because their OAHI was above 1/h. Thus, the PS group comprised 17 children (8 girls and 9 boys). The inclusion criteria for the control group were snoring free at home (never snored or 1-2 nights a month), PSG snoring time $< 1\%$ and OAHI < 1/h. Eight control children were excluded from the final analyses; five had snoring time over 1% and three had an OAHI over 1/h. Thus, the control group comprised 27 children (13 girls and 14 boys).

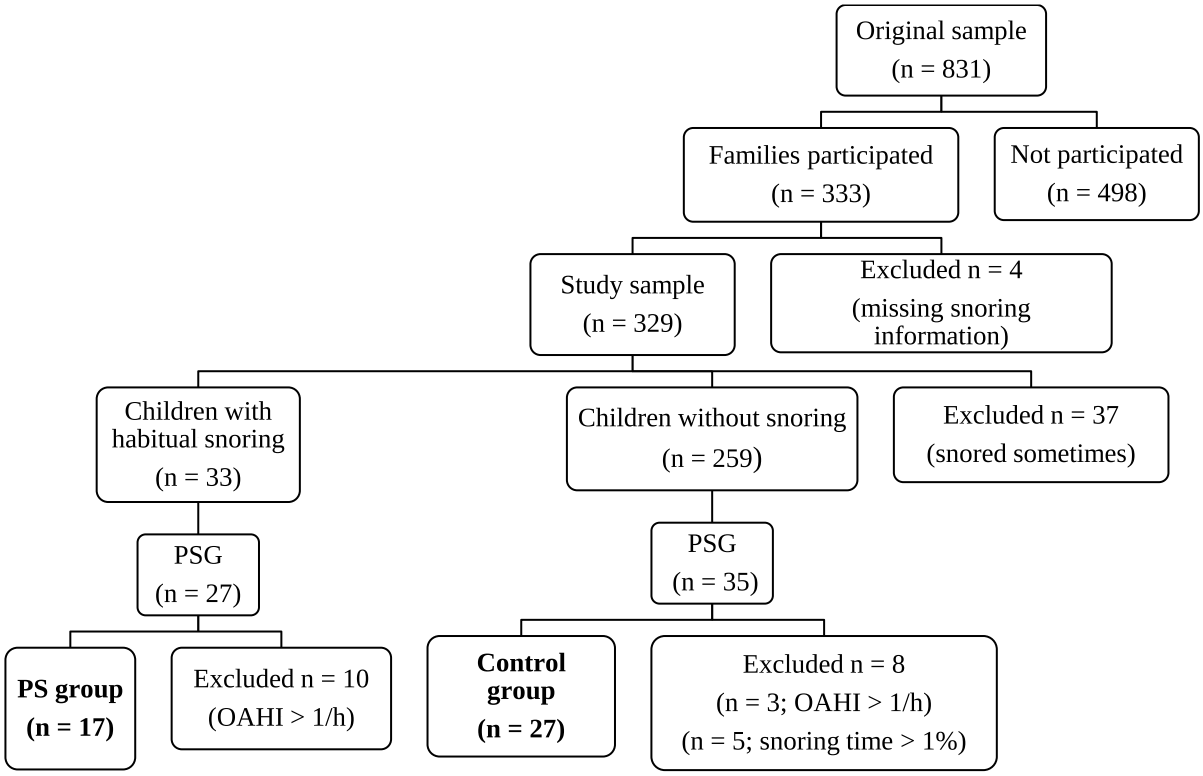

Fig. 1. Sampling procedure. PSG, polysomnography; OAHI, obstructive apnea-hypopnea index

Measurements of Behavioral Problems and Neurocognitive Functioning

Behavioral Assessment. Parents completed the questionnaires during their visit to the Sleep Laboratory and teachers at school. Behavioral problems were assessed using the Problem Scales of the Child Behavior Checklist (CBCL, parent version) and equivalent Teacher Report Form (TRF) (Achenbach & Rescorla, 2001), both for children aged 6 to 18 years. The questionnaires yield 8 scales: Anxious/Depressed, Withdrawn/Depressed, Somatic Complaints (these three constitute an index for Internalizing Problems), Social Problems, Thought Problems, Attention Problems, Rule-Breaking Behavior, and Aggressive Behavior (the last two constitute an index for Externalizing Problems), and Total Problems. The CBCL and TRF indices have a mean *T*-score of 50 and a standard deviation of 10. *T*-scores of 60 – 63 are considered as cut-off scores for borderline and $T > 63$ for clinical behavioral problems (Achenbach & Rescorla, 2001). The Conners' Parent Rating Scale-Revised (CPRS-R) and the

Conners' Teacher Rating Scale-Revised (CTRS-R) (Conners, 1997), both for children aged 3 to 17 years, were used to identify attentional problems. The CPRS-R and CTRS-R yield seven attention indices: Attention Deficit Hyperactivity Disorder (ADHD) Index, Global Index: Restless-Impulsive, Global Index: Emotional Lability, Global Index: Total Index, DSM-IV: Inattentive, DSM-IV: Hyperactive-Impulsive, and DSM-IV: Total. The CPRS-R and CTRS-R indices have a mean *T*-score of 50 and a standard deviation of 10. The scores were considered borderline when *T*-score is between 56 – 60. *T*-scores of 61 – 65 indicate a possible significant problem, and *T*-scores > 65 indicate a clinically significant problem (Conners, 1997).

Neuropsychological assessment. All children underwent a three-hour neuropsychological evaluation at the Sleep Laboratory. A trained psychologist or a trained postgraduate student (in psychology) administered the standardized tests to each child individually. The examiner was unaware of the child's SDB status. The intellectual functioning of the children was evaluated using the Finnish version of the Wechsler Intelligence Scale for Children (WISC-III) (Wechsler, 1999). Verbal Intelligence Quotient (VIQ), Performance Intelligence Quotient (PIQ), and Full Scale Intelligence Quotient (FSIQ) were estimated according to the manual using six subtests (Information, Similarities, Arithmetic, Picture Completion, Block Design, and Object Assembly). Core neurocognitive functions were assessed with the Developmental Neuropsychological Assessment for children aged 3 to 12 years (NEPSY, Finnish version) (Korkman, Kirk, & Kemp, 2000). The five age-appropriate domains (and their subtests) were as follows: Attention and Executive Function (Tower, Auditory Attention and Response Set, Visual Attention), Language Function (Phonological Processing, Comprehension of Instructions, Speeded Naming), Sensorimotor Function (Fingertip Tapping, Imitating Hand Positions, Visuomotor Precision), Visuospatial Function (Design Copying, Arrows), and Memory and Learning Function (Memory for Faces, Memory for Names, Narrative Memory).

Statistical Analysis

PSG, neurocognitive, and behavioral parameters were compared between the groups. In addition, correlations between the respiratory parameters and neurobehavioral variables were calculated for all the children (combined groups). The statistical analyses were conducted using Statistical Package for the Social Sciences (SPSS) software (Version 25.0 for Mac OS X). Nonparametric tests were chosen because some of the parameters were not normally distributed and the sample sizes were small. Pearson Chi-square analyses with Fisher's exact test values were used to compare the frequencies of parents' education and combined borderline and clinical behavioral impairments between the study groups. The remaining demographic data, sleep and respiratory parameters, and neurobehavioral scores between the two groups were tested using the Mann-Whitney *U*-test. For a description of the data, the raw scores of the CBCL, TRF, CPRS-R, and CTRS-R were individually converted into *T*-scores and into indices. Spearman correlation coefficients were calculated for the combined groups between neurobehavioral outcomes (VIQ, PIQ, FSIQ, the five NEPSY domains, CBCL, and CPRS-R indices) and all the sleep variables. Effect sizes for Mann-Whitney *U*-tests were calculated from the formula: $r = \frac{Z}{\sqrt{N}}$ (Fritz, Morris, & Richler, 2012) and for Chi-square analyses $r = \sqrt{\frac{x^2(1)}{N}}$ (Rosenthal & DiMatteo, 2001). The effect size of .10 indicates small effect, .30 medium effect, and .50 large effect. Statistical significance was set at $p < 0.05$.

Results

Demographic Characteristics and PSG Parameters

The PS and control groups did not differ in gender, age, body mass index, or in parental education level (Table 1)**.** Parents reported that children with PS had more daytime sleepiness than control children ($U = 124.50$, $p = .01$, $r = -.39$).

Table 1. Demographic data of the PS and control groups.

	PS (N = 17)	Control (N = 27)	p
Gender, males (%)	9 (53)	14 (52)	.944
Age (years)	8.1 ± 1.0	8.7 ± 1.1	.165
Body mass index	22.8 ± 2.5	22.1 ± 3.9	.329
Maternal education	N = 16	N = 27	.698
Basic (%)	0 (0)	1 (4)	
Vocational training (%)	7 (44)	14 (52)	
High school/College (%)	5 (31)	5 (18)	
University (%)	4 (25)	7 (26)	
Paternal education	N = 16	N = 27	.375
Basic (%)	3 (19)	2 (8)	
Vocational training (%)	2 (13)	9 (33)	
High school/College (%)	4 (25)	7 (26)	
University (%)	7 (44)	9 (33)	

As expected, the PS group snored more than the control group (U = .000, p < .001, r = -.85, Table 2). In general, the groups did not differ by sleep architecture parameters, but the PS group had more arousals than the control group (U = 141.00, p =.033, r = -.32). The number of obstructive breathing events and desaturations were similar between the groups, but the PS group had significantly more flow limitation (U = 99.00, p < .001, r = -.55), and periods with increased diaphragmatic EMG (U = 170.00, p =.016, r = -.36). $TcCO_2$-values were normal in all children (highest values < 6 kPa).

Table 2. Sleep architecture and respiratory characteristics of the PS and control groups.

PSG variables	PS (N = 17)	Control (N = 27)	p
Snoring time (%)	8.1 ± 5.5	0.13 ± 0.2	< .001
TST (min)	546.4 ± 36.1	541.2 ± 29.5	.500
SE (%)	94.1 ± 3.2	93.4 ± 4.0	.571
REM (%)	17.7 ± 4.1	17.1 ± 3.8	.539
N1 (%)	5.1 ± 2.2	5.8 ± 3.4	.579
N2 (%)	41.4 ± 9.3	42.7 ± 6.6	.971
N3 (%)	35.9 ± 8.6	34.5 ± 7.6	.885
OAHI/h	0.2 ± 0.2	0.2 ± 0.2	.843
ARI/h	8.9 ± 2.5	7.5 ± 3.3	.033
SpO_2 (%)	97.9 ± 0.6	97.6 ± 0.6	.168
SpO_2 < 90%	0.4 ± 1.0	0.2 ± 0.6	.689
FL (%)	2.8 ± 3.2	0.5 ± 1.3	< .001
D-EMG (%)	0.7± 1.8	0.03 ± 0.2	.016
ODI3/h	0.2 ± 0.3	0.3 ± 0.3	.167

Data presented as M ± SD. PSG, Polysomnography; TST, Total Sleep Time; SE, Sleep Efficiency; REM, Rapid Eye Movement Sleep; N1, Stage 1 Non-REM Sleep; N2, Stage 2 Non-REM Sleep; N3, Stage 3 Non-REM Sleep; OAHI, Obstructive Apnea Hypopnea Index; ARI, Arousal Index; SpO_2, Mean Oxygen Saturation; SpO_2 < 90%, minutes per h of TST with SpO_2 less than 90%; FL, Flow Limitation; D-EMG, Increased diaphragmatic Electromyography; ODI3, Oxygen Desaturation Index, number of desaturations ≥ 3% per hour of TST.

Behavioral Problems. The CBCL questionnaires were obtained from 17 children with PS and 26 controls and the TRF from 14 children with PS and 25 control children. The parents of the PS group reported more anxious/depressed symptoms (U = 103.50, p = .002, r = -.48), withdrawn/depressed symptoms (U = 105.50, p = .004, r = -.44), somatic complaints (U = 140.50, p = .043, r = -.31), thought problems (U = 77.00, p < .001, r = -.56), and social problems (U = 133.50, p = .028, r = -.34). The children with PS showed more parent-reported internalizing (U = 99.50, p = .002, r = -.46) and total symptoms (U = 100.00, p = .003, r = -.46) than the controls.

As presented in Figure 2, the PS group displayed proportionally more borderline or clinically significant impairments (T-score ≥ 60) in Internalizing ($\chi^2(1)$ = 10.90, p = .001, r = .50), Externalizing ($\chi^2(1)$ = 5.13, p = .024, r = .35), and Total ($\chi^2(1)$ = 13.90, p < .001, r = .57) Problem scores compared with controls. Approximately half of the children with PS were within borderline or clinical range scores on the CBCL Internalizing, Externalizing, and Total indices compared with 12, 15, and 4% in controls.

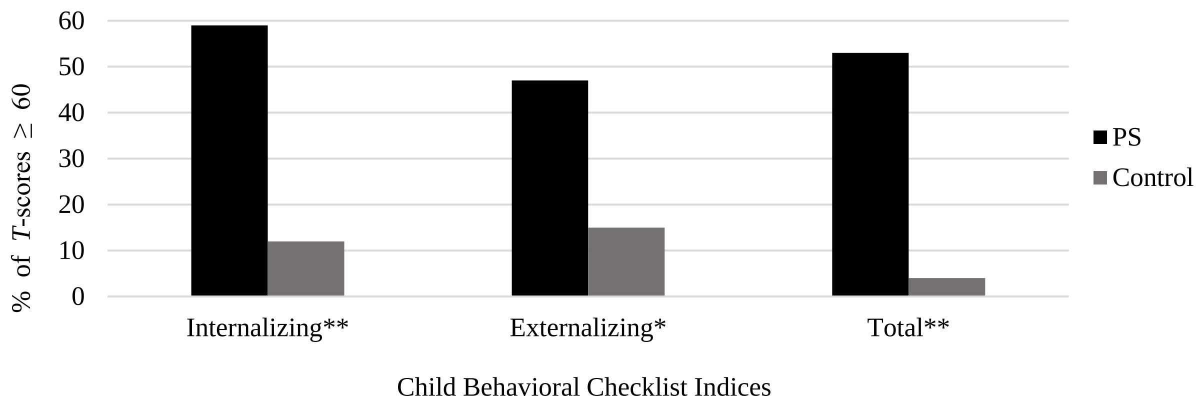

Fig. 2. The percentage of children with borderline/clinical scores on the CBCL indices. * p < 0.05, ** p < 0.01

Further, compared with controls, the parents of the PS group reported their children had more attentional problems (Fig. 3), both inattentive (DSM-IV Inattentive: U = 122.50, p = .014, r = -.38) and hyperactive-impulsive behavior (ADHD Index: U = 135.50, p = .033, r = -.33; Global Index: Total: U = 140.00, p = .044, r = -.31). The children with PS had more borderline or clinically significant impairments in ADHD Index ($\chi^2(1)$ = 9.46, p = .002, r = .47), Global Index: Restless-Impulsive ($\chi^2(1)$ = 9.46, p = 002, r = .47), DSM-IV: Inattentive ($\chi^2(1)$ = 9.46, p = .002, r = .47), and in DSM-IV: Total ($\chi^2(1)$ = 6.81, p = .009, r = .40).

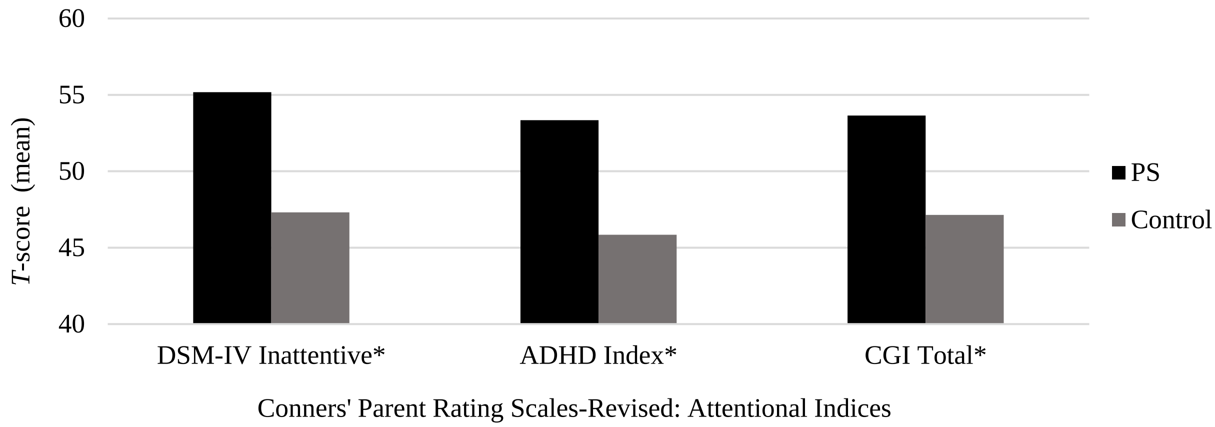

Fig. 3. The scores on the CPRS-R indices for the PS and control groups. DSM-IV, Diagnostic and Statistical Manual of Mental Disorders; ADHD, Attention Deficit Hyperactivity Disorder; CGI; Conners' Global Index.
* $p < 0.05$, ** $p < 0.01$

Teachers did not report any differences between the two groups in any of the TRF or CTRS-R scales, and all of the indices and scales for the PS group reported by teachers were within the normal range.

Neurocognitive Functions. On average, the VIQ, PIQ, and FSIQ scores were within the normal range for both groups as well as the core neurocognitive functions measured with the NEPSY (Table 3). No significant differences were found between the PS and control groups in the IQ scores or the core neurocognitive functions. However, the children with PS performed significantly worse than the control children in one visuospatial subtest of the NEPSY, i.e., Arrows (U = 145.00, p = .039, r = -.31).

Table 3. Scores for neurocognitive functions for the PS and control groups.

	PS group ($N = 17$) Mean ± SD (Range)	Control group ($N = 27$) Mean ± SD (Range)	p
WISC-III			
Verbal IQ	99.8 ± 12.3 (73-120)	107.6 ± 17 (73-156)	.125
Performance IQ	104.2 ± 16.8 (81-136)	108.3 ± 18.4 (71-132)	.365
Full Scale IQ	101.7 ± 13.1 (80-123)	106.8 ± 16.6 (79-141)	.257
NEPSY			
Attention and Executive Function			
Tower	12.2 ± 2.3 (8-15)	11.3 ± 3.0 (3-16)	.337
Auditory Attention and Response Set	9.5 ± 4.1 (2-17)	10.7 ± 3.2 (3-15)	.264
Visual Attention	13.4 ± 3.9 (7-19)	11.9 ± 3.7 (3-19)	.231
Language Function			
Phonological Processing	11.4 ± 2.1 (6-15)	11.3 ± 2.9 (3-16)	.874
Comprehension of Instructions	10.1 ± 3.1 (3-15)	10.0 ± 3.7 (3-16)	.903
Speeded Naming	10.2 ± 2.9 (6-16)	10.4 ± 3.3 (5-21)	.865
Sensorimotor Function			
Fingertip Tapping	13.0 ± 3.4 (1-16)	13.0 ± 1.7 (10-16)	.331
Imitating Hand Positions	9.4 ± 1.5 (7-13)	10.0 ± 2.3 (5-14)	.149
Visuomotor Precision	8.1 ± 3.0 (3-14)	8.5 ± 4.4 (1-17)	.799
Visuospatial Function			
Design Copying	10.4 ± 3.0 (1-14)	10.7 ± 2.3 (7-16)	.961
Arrows	9.6 ± 3.2 (3-13)	11.6 ± 2.6 (7-16)	.039*
Memory and Learning Function			
Memory for Faces	10.6 ± 4.0 (2-15)	9.5 ± 3.7 (1-15)	.235
Memory for Names	9.9 ± 3.0 (5-15)	9.3 ± 2.5 (5-15)	.497
Narrative Memory	10 ± 3.7 (1-16)	10.1 ± 3.4 (2-15)	.942

* $p < 0.05$, ** $p < 0.01$

Associations Between Daytime Sleepiness, Respiratory, and Neurobehavioral Variables

Correlations calculated for the combined groups yielded some significant associations (Table 4). Daytime sleepiness was consistently associated with behavior and attention, but in neurocognitive functions only with Tower. Snoring time was associated with behavior and attention: children with snoring had more internalizing, total, and attentional (GCI: Total, DSM-IV: Inattentive, and DSM-IV: Total) problems. Neurocognitive functions were related to two respiratory variables: flow limitation was associated with reduced scores in Auditory Attention and Response Set, and children with higher oxygen desaturation index had reduced Speeded Naming and Comprehension of Instructions. Mean oxygen saturation was associated with Comprehension of Instructions and children with more time with oxygen saturation < 90% had more problems with Visual Attention. Other respiratory variables (OAHI and EMG%) did not correlate with any of the neurobehavioral variables.

Table 4. Spearman's correlation coefficients for combined groups between daytime sleepiness, respiratory, and neurobehavioral variables

	Daytime sleepiness	Snoring time	FL%	ODI3	OAHI	SpO_2	SpO_2 <90%	EMG%
CBCL								
Anxious/Depressed	.444**	.395**	ns	ns	ns	ns	ns	ns
Withdrawn/Depressed	.368*	.307**	ns	ns	ns	ns	ns	ns
Somatic Complaints	.310*	ns	ns	ns	ns	ns	ns	ns
Social Problems	.510**	.351*	ns	ns	ns	ns	ns	ns
Thought problems	.483**	.424**	ns	ns	ns	ns	ns	ns
Attention Problems	.513**	.368*	ns	ns	ns	ns	ns	ns
Rule-Breaking Problems	ns	ns	ns	ns	ns	ns	ns	ns
Aggressive Behavior	.394**	ns	ns	ns	ns	ns	ns	ns
Internalizing Problems	.424**	.369*	ns	ns	ns	ns	ns	ns
Externalizing Problems	.371*	ns	ns	ns	ns	ns	ns	ns
Total Problems	.501**	.380*	ns	ns	ns	ns	ns	ns
CPTR-S attention Indices								
Attention Deficit Hyperactivity Disorder (ADHD) Index	.424**	ns	ns	ns	ns	ns	ns	ns
GI: Restless-Impulsive	.496**	ns	ns	ns	ns	ns	ns	ns
GI: Emotional Lability	.499**	ns	ns	ns	ns	ns	ns	ns
GI: Total Index	.530**	.311*	ns	ns	ns	ns	ns	ns
DSM-IV: Inattentive	.443**	.355*	ns	ns	ns	ns	ns	ns
DSM-IV: Hyperactive-Impulsive	.422*	ns	ns	ns	ns	ns	ns	ns
DSM-IV: Total	.457**	.305*	ns	ns	ns	ns	ns	ns
WISC-III								
Verbal IQ	ns	ns	ns	ns	ns	ns	ns	ns
Performance IQ	ns	ns	ns	ns	ns	ns	ns	ns
Full Scale IQ	ns	ns	ns	ns	ns	ns	ns	ns
NEPSY								
Tower	.317*	ns	ns	ns	ns	ns	ns	ns
Auditory Attention and Response Set	ns	ns	-.412**	ns	ns	ns	ns	ns
Visual Attention	ns	ns	ns	ns	ns	ns	-.308*	ns
Phonological Processing	ns	ns	ns	ns	ns	ns	ns	ns
Comprehension of Instructions	ns	ns	ns	-.384*	ns	.333*	ns	ns
Speeded Naming	ns	ns	ns	-.308*	ns	ns	ns	ns
Fingertip Tapping	ns	ns	ns	ns	ns	ns	ns	ns
Imitating Hand Positions	ns	ns	ns	ns	ns	ns	ns	ns
Visuomotor Precision	ns	ns	ns	ns	ns	ns	ns	ns
Design Copying	ns	ns	ns	ns	ns	ns	ns	ns
Arrows	ns	ns	ns	ns	ns	ns	ns	ns
Memory for Faces	ns	ns	ns	ns	ns	ns	ns	ns
Memory for Names	ns	ns	ns	ns	ns	ns	ns	ns
Narrative Memory	ns	ns	ns	ns	ns	ns	ns	ns

FL, Flow Limitation; ODI3, Oxygen Desaturation Index, number of desaturations ≥ 3% per hour of TST; OAHI, Obstructive Apnea Hypopnea Index; SpO_2, Mean Oxygen Saturation; SpO_2 < 90%, minutes per h of TST with SpO_2 less than 90%; EMG, Increased diaphragmatic Electromyography * $p < 0.05$, ** $p < 0.01$

Discussion

To the best of our knowledge, the present study is the first work that evaluates cognition and behavioral performance in school-aged children who snore employing the suggested definition for PS (Kaditis et al., 2016) with quantified snoring time. In this way, our study makes an important contribution to the current literature regarding the relationship between snoring and neurobehavioral outcomes. As such it raises awareness among clinicians that SDB without OSA, namely PS in children, may affect children's daytime functioning and may have long-term effects on their social and academic competence.

The PS and control groups had similar demographic characteristics except for more daytime sleepiness reported in children with PS. The only parameter describing sleep architecture that differed between the PS and control group was the arousal index, although the number of arousals remained low and within normal values (Tapia et al., 2008) in both groups. Hence, our finding is in line with previous studies, where sleep architecture disruption has been found to be uncommon in snoring (Bourke et al., 2011b; Zhu et al., 2014). Due to the inclusion criteria, the PS group presented with more snoring than the control group, but other conventional respiratory parameters did not differ between the groups. However, the PS group had more partial upper airway obstruction (flow limitation and increased diaphragmatic EMG). This is a common clinical finding in children's sleep recordings (see Norman, Pithers, Teng, Waters, & Sullivan, 2017). In adults, prolonged partial obstruction has been found to be associated with depressive symptoms and decreased quality of life (Anttalainen et al., 2016), but a corresponding evaluation has not previously been performed in children.

The study confirmed, as expected, that the parents of children who experience primary snoring report a higher frequency of internalizing and externalizing behaviors, and more attention problems and ADHD symptoms than parents of children without snoring. Teachers did not report differences in behavioral measures between children with and without snoring.

Even though behavior problems were expected, the percentages for behavioral problems in the PS group were surprisingly markedly elevated compared with the expected level of impairment (16%) in a normal child population (Achenbach & Rescorla, 2001). In particular, the children with PS had higher levels of parent-reported anxious, depressed, thought, and psychosomatic symptoms, and more social problems than the non-snoring controls. Consistent with the current findings, internalizing symptoms have been reported earlier with both school-aged children (Beebe et al., 2010; Bourke et al., 2011b; O'Brien et al., 2004) and preschoolers with PS (Jackman et al., 2012).

Low parent-teacher agreement on behavioral symptoms was partly expected (Arman et al., 2005; Kohler et al., 2013; O'Brien et al., 2011), and is consistent with earlier studies (Narad et al., 2015). This inconsistency between parent and teacher ratings may be related to the different expectations towards children's behavior at home and in school. Low multi-informant agreement may also reflect variations in functioning across situations (Achenbach, McConaughy, & Howell, 1987). For example, attention may fluctuate during the day or children may be tired after an intensive school day, which in turn may increase impulsive and restless behavior in the afternoon and evening. Therefore, parents are more likely to observe certain behaviors than teachers. Some parents may have also been aware of the association between snoring and behavioral problems and this might have affected their evaluations.

An unexpected finding in our study with verified snoring was that the neurocognitive findings remained minor, and no diffuse pattern of impairments was found. No differences between children with PS and controls were found, except for the poorer performance of children with PS in one visuospatial subtest. In addition to the visual spatial component (such as judgement of line orientation) of the subtest Arrows, it requires selective visual attention and impulse inhibition. This finding may be an artifact related to large variance in the study group or alternatively related to attention problems. The result may also be explained by a normal variance in a battery of tests or the likelihood of one significant outcome with alpha inflation. Our finding is concurrent with some earlier studies (Calhoun et al., 2009; Jackman et al., 2012), and suggests that neurocognitive deficits may not be as common in children with PS as previously suggested. Similarly, another study with verified and quantified snoring did not find differences in memory consolidation between the children with PS and healthy controls either (Maski et al., 2017).

Definite explanations for higher behavioral problems cannot be determined based on the results of this study. Most likely, the associations between PS and neurobehavioral outcomes are multifactorial and intercorrelated. However, the study revealed consistent associations between parent-reported behavior, attention problems, and daytime sleepiness. Possibly, daytime sleepiness is a modifying factor between PS and behavior; either behavioral problems and psychosomatic complaints may be further increased with daytime sleepiness, or they may be symptoms of daytime sleepiness related to PS. In some children, behavioral problems could also be attributed to comorbid attention problems as demonstrated in children with attention-deficit/hyperactivity disorder. Comorbidity between attention deficits and behavioral problems is a rather well-established finding (Lonigan et al., 2017). Externalizing disorders are reported to be most prevalent in children with ADHD, but approximately 25% of children with ADHD exhibit an anxiety disorder (see Jarrett & Ollendick, 2008).

The results of our study highlight the potential impact of PS on internalizing symptoms, attentional control and behavioral regulation. The results confirmed that pediatric SDB patients have an increased risk of developing internalizing (Beebe et al., 2010; Bourke et al., 2011b; O'Brien et al., 2004) and attentional problems, with both inattentive and hyperactive-impulsive presentation (see Biggs et al., 2014; Sedky, Bennett, & Carvalho, 2014). Also, Chervin, Ruzicka, Hedger Arcbold, and Dillon (2005) concluded that snoring, especially if left untreated, is a contributing factor in the development of hyperactivity. Moreover, it has been suggested (Smith et al., 2017) that possible alterations in sleep structure in PS may affect the underlying neural mechanisms that support both the directed and sustained attention and fronto-striatal circuits related to behavioral and emotional regulation.

Daytime sleepiness was consistently associated with behavior and attention, but not with neurocognitive functions. Because the snoring time was quantified in our study, the results add to the present body of knowledge by presenting that an increasing amount of snoring increases both behavioral problems and attention in dose response manner. Snoring per se did not correlate with cognitive measures. The share of prolonged partial obstruction (FL%), however, was negatively associated with auditory attention. In adults, prolonged partial obstruction is found to induce daytime sleepiness (Pelin, Karadeniz, Ozturk, Gozukirmizi, & Kaynak, 2003). The consequences of prolonged partial obstruction have not previously been evaluated in children. It is possible that snoring with prolonged partial obstruction results in daytime symptoms and weakens selective and sustained auditory attention. Future studies with larger sample sizes are therefore needed to ascertain whether snoring with prolonged partial obstruction is more severe than snoring without partial obstruction.

Clinical Implications and Conclusion

There are several strengths in the present study; first and foremost being the fact that we quantified the amount of snoring. Children underwent a comprehensive neuropsychological assessment of core neurocognitive functions. In addition, we used both parent- and teacher-reported, well-validated and widely used questionnaires to get a broad picture of the children’s behavior in everyday life. The current study also has limitations. Forty percent of the families returned the sleep questionnaire. The return rate of the forms was somewhat lower than in other pediatric sleep studies (Arman et al., 2005; Brockmann et al., 2012; O’Brien et al., 2004). This rather poor return rate is partly related to methodological choices of the study; the questionnaire was handed out during physical examination by the school nurse or in class by the teacher and parents filled out the form at home. Therefore, the researchers were unaware of families first receiving the questionnaires and thus unable to send reminders for participation. The participation rate to the clinical part of the study was quite low, probably due to the inconvenience of the study protocol. During the PSG studies, the children (and most parents) slept two successive nights at the sleep laboratory, and on the day of the psychological assessment the children were absent from school. Because of the small sample size, we decided not to match the two study groups precisely to age and gender.

Despite the limited number of participants in our study, the results make an interesting contribution by indicating that school-aged children with snoring may have not only snoring but also prolonged partial obstruction. PS is currently considered to be the mildest form of SDB even though children with PS experience similar cognitive and even elevated behavioral deficits compared with children with OSA. It is possible that snoring represents a unique and complex phenotype of SDB (Biggs et al., 2014). We therefore suggest that snoring with and without increased respiratory efforts should be taken into account in future studies. For scientific purposes, parent and teacher ratings provide an opportunity to assess children's behavior, attention, and executive functions in natural situations in contrast to psychological tests conducted in clinical environments.

Pediatric snoring is common, and it is important therefore to raise awareness that children with snoring are at risk for behavioral and attentional problems. The fact that the majority of children with PS in this study had borderline or clinically significant behavioral problems is a major concern. If left untreated, these difficulties may adversely impact learning and lead to mental problems, social isolation, and poor school performance. Clinicians evaluating children with snoring should be aware of the comorbid behavioral risk factors. The possibility of sleep disorder should be kept in mind when assessing children with behavioral problems or attentional deficits. A comprehensive psychological assessment including both psychological testing and multi-informant behavioral ratings is therefore recommended as a standard procedure.

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