Sleep-wake schedules in preschool children who snore

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ABSTRACT

Purpose: The aim of the study was to compare sleep-wake schedules between snoring and nonsnoring preschool age children.

Material and Methods: Daytime and nighttime sleep duration, daytime and nighttime symptoms were assessed in 34 children at preschool age who snore (5.38 ± 1.21 years) and in 66 age- and sex-matched nonsnorers (5.67 ± 1.12 years). The snoring group consisted of children with obstructive sleep apnea (OSA) scores <+3.5 and > -1, the nonsnoring group with OSA score <-1.

Results: Children who snore differ from the nonsnorers in daytime sleep duration (51.62 ± 28.9 minutes vs. 10.70 ± 20.2 minutes; p<0.001), but not in nighttime sleep (10.97 ± 0.52 hours vs. 9.83 ± 1.34 hours; p>0.05). The percentage of children with daytime napping was higher in the snoring group than in the nonsnorers (47.1% vs. 9.1%; p<0.00004), and parents-reported behavioral problems were more frequent in children who snore (41.2% vs. 19.7%; p<0.02). Multivariate odds ratios, including variables for nighttime (sleep apnea) and daytime symptoms (daytime napping and oral breathing), showed that regular sleep during the day was the most predictive of snoring (OR=6.1; 95%CI 1.76-21.04; p<0.005).

Conclusion: In preschool age children, when the daytime nap begins to disappear, snoring may have an effect on daytime schedule through an increased need for daytime sleep.

Key words: snoring, sleep habits, daytime sleep, sleep-disordered breathing (SDB), behavioral problems

INTRODUCTION

In the last years, views on the consequences of snoring in children have undergone a change. Habitual snoring (snoring on most nights) can be associated with adverse effects, even if it does not lead to gas exchange disturbances or sleep disruption (primary snoring) [1]. Primary snoring is diagnosed in the absence of apnea, hypoventilation and hypoxemia in polysomnography in contrast to obstructive sleep apnea hypopnea syndrome (OSAHS). The clinical course of primary snoring is mild; however, according to recent psychological research studies, snoring is related to an increased risk factor for behavioral disorders, poor school performance and mood disturbances (anxious/depressed mood) [2,3]. One of possible explanations is the effect snoring has on the quantity or quality of sleep.

Snoring is a measure of partial upper airway occlusion, an indicator of increased upper airway resistance and one of

the symptoms of disordered breathing during sleep [4]. Sleepdisordered breathing (SDB) includes: primary snoring, upper airway resistance syndrome (UARS) and OSAHS [5]. Primary snoring has been reported in 10% of children, 1-3% of children are likely affected by OSAHS. Untreated SDB can have many consequences, including growth abnormalities, cardiovascular remodeling, neurocognitive deficits, behavioral disorders (depression, anxiety, Attention Deficit Hyperactivity Disorder (ADHD)) and social problems [6,7]. Snoring manifests in children of all ages, those "healthy" and ill: during the course of upper respiratory tract diseases, local or general disorders, e.g. neuromuscular diseases, genetic diseases, craniofacial anomalies, allergies, and obesity [8].

The age regarded as at the highest risk of snoring occurrence in children ranges from 4 to 10 years, at a time when adenotonsillar size is maximal relative to the airway. This is also the time of child's simultaneous development in many areas: forming social relations, maturation towards school readiness, and intensive mental effort. It is also a time of a reduced need for daytime sleep in the population of healthy children, as only in this way so many developmental tasks can be accomplished. [9]. Poor quality sleep and quantitative sleep disorders in children with OSAHS are commonly known.

The aim of the current study was to assess sleep duration and napping habits in a sample of snoring and nonsnoring children at preschool age.

MATERIAL AND METHODS

Patient selection. The snoring group consisted of preschool children aged 3-7 years (in Poland children start school at the age of 7) admitted to our Sleep Laboratory between March 2006 and September 2008. Children who snore were recruited from among patients referred to our Department by family doctors or pediatricians for differential diagnosis of sleep-disordered breathing. The control group of age- and sex-matched healthy children who do not snore were recruited randomly from 4 kindergartens located in university town. The patients were allocated in the study group (children who snore) or control group (nonsnorers) based on the obstructive sleep apnea (OSA) score described by Brouillette et al [10]. OSA score was calculated according to the equation with three variables - symptoms of SDB: OSA score = 1.42D+1.41A+0.71S-3.83 (D means breathing difficulty, A is apnea during sleep, S is snoring). Values assigned to D and S were 0-never, 1-occasionally, 2-frequently, 3-always. Values assigned to A were 0-No, 1-Yes. OSA scores < -1 indicate absence of OSA, OSA scores between -1 and 3.5 may or not may indicate OSA, OSA score >3.5 is "diagnostic" of OSA (OSA score is based on clinical history only and cannot be used for real OSA identification - polysomnographic examination fulfils this role) [11]. In our study, the snoring group consisted of children with OSA scores <3.5 and >-1, and the non-snoring group included children with OSA scores < -1. Children with the highest OSA scores (> +3.5), as the group of the highest risk of OSA, and children with history of chronic genetic or neurologic disease were excluded.

Sleep habits and clinical history. Structured interviews were performed for 3 various sleep habits – A, B, C according to Iglowstein study [9]: A. Bedtime - When does the child usually go to bed? In response, give full hour or hour and 30'. B. Wake time - When does the child usually wake up? In response, give full hour or hour and 30'. C. Daytime napping habits - How long does the child sleep during daytime? In response, give the number of minutes: 0', 15', 30', 45', 60', 75', 90', 105', 120'. Nighttime sleep duration was calculated from bedtime and wake time.

Daytime sleepiness (hypersomnia or a reduced latency to sleep during daytime) was assessed in 4 items characteristic of daytime sleep/sleepiness: HOW – what is sleep onset like, WHERE – the place of falling asleep, WHEN – the time of falling asleep and SLEEPY signs in child's behavior: Does your child: a. unintentionally fall asleep during the day (e.g. during play time) (Yes-1, No-0); b. fall asleep elsewhere (e.g. outside his/her bed, on the carpet, at the table) (Yes-1, No-0); c. fall asleep apart from regular nap time (Yes-1, No-0); d. often demonstrate behaviors indicating somnolence e.g. yawning, rubbing the eyes, eye dubbing (Yes-1, No-0)? Daytime sleepiness was recognized based on two or more positive answers in parents-reported subjective evaluation.

Behavioral ADHD-related problems were investigated with 6 questions in 3 categories: inattention (3-items), impulsivity (2-items) and hyperactivity (1-item), worked out according to ADHD criteria by DSM-IV 4th ed [12]. Does your child often: - leave seat in situations in which remaining seated is expected? - have difficulty awaiting turn? - interrupt or intrude on others? - lose things necessary for tasks or activities? - fail to finish his/her play or work? - fail to give close attention to details in work or other activities? Children with 3 or more positive answers given by parents were coded as suspected to have behavioral problems (Yes-1) and clinical psychologist consultation was offered. Those with <3 positive answers were coded as without behavior problems (No-0). The screening result was not treated as ADHD diagnosis.

Additionally, home and family environment were compared: family model (parents and the number of children in the family), place of sleep (child's own bedroom), history of bedwetting, exposure to pets and smoking ("Is your child exposed to indoor smoking?"), parents' history of habitual snoring. To assess the child's personal medical history we asked the parents whether their child had ever been diagnosed with and/or treated for a. adenotonsillar hypertrophy, b. upper airway infections occurring more frequently than 7 times per year, c. gastroesophageal reflux disease, d. allergic disease (d1.of skin, d2. of alimentary tract, d3 of respiratory tract). All the questionnaires were completed by one of two interviewers (JW, MKK). Body mass index (BMI) was calculated as the ratio between weight and height squared (kg/m²). Children with BMI of 90th percentile were classified as overweight and children with BMI above 97th percentile were referred to as obese.

The study project was approved by the local Bioethical Committee of the Medical University of Bialystok.

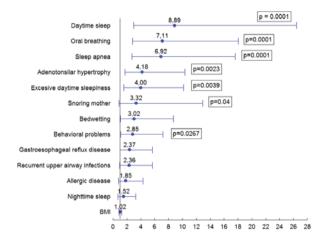
Statistical analysis

In statistical analysis, the Student's t-test was applied to compare variables of a normal distribution (BMI), the Mann-Whitney *U*-test to compare variables of non-parametric distribution (age), the Shapiro-Wilk test to verify the statistical shape of tested variable distribution (BMI and age), as well as the chi-square test for independence, and the Yates' chi-square test to compare qualitative and categorized variables. In order to evaluate the relationship between independent variables (risk factors) and dichotomous dependent variable (snoring occurrence in children), logistic regression was employed. All the potential risk factors of snoring occurrence among

Figure 1. Histogram of OSA Score (the snoring group consisted of children with OSA scores <+3.5 and >-1, and the nonsnoring group consisted of children with OSA score <-1).



Figure 2. Predictive factors of snoring in preschool age children.



children were the subject of analysis. In order to indicate the strongest and independent factors of snoring occurrence among children, all the independent variables, significantly changing the compatibility of one-factor models, were included in the multi-factor model. For all the statistical tests used, α =0.05 level of relevance was accepted. The statistical analysis was carried out by means of the STATISTICA 8.0 program.

RESULTS

The snoring group consisted of 34 children at the age of 3 years 6 months to 7 years (5.38±1.21 years) and the control group included 66 healthy children at the age of 3 years 2 months to 7 years 5 months (5.67 ± 1.12 years). The mean OSA score in the snoring group was 1.18±0.86 (median 1.13) and in 91.2% (31/34) did not exceed +2.5, whereas the mean OSA score in the nonsnorers was -2.16±0.57 (median -1.71) (Fig. 1); 7 children with OSA score \geq +3.5 were excluded.

No significant differences were found for age, gender, place of residence, BMI, number of siblings, history of bedwetting, father's snoring or home environment between the snoring and control group (Tab. 1). Children who snore differed from the non-snoring group in daytime sleep duration (51.62±28.9 minutes vs. 10.70±20.2 minutes; p<0.001), but not in nighttime sleep (10.97±0.52 hours vs. 9.82±1.34 hours; p>0.05). The percentage of children with daytime napping was higher in the snoring group than in the nonsnorers (47.1% vs. 9.1%; p<0.00004) and behavioral problems reported by parents were more frequent in children who snore (41.2% vs. 19.7%; p<0.02). Snoring was not related to BMI, personal medical history, home environment, or family structure.

In univariate analysis, the most typical factors for snoring were: regular daytime sleep (OR=8.89; 95%CI 2.99-26.43; p<0.0001) (Fig. 2), daytime oral breathing (OR=7.1; 95%CI 2.80-18.06 p<0.001), sleep apnea (OR= 6.9; 95% CI 2.72-17.59; p<0.001), adenotonsilar hypertrophy (OR=4.2; 95%CI 1.69-10.35; p<0,002), daytime sleepiness (OR=4.0; 95% CI 1.58-10.14; p<0,0039) and maternal snoring (OR=3.0; 95% CI 1.05-8.70; p<0.04) (Tab. 2).

Multivariate odds ratios, including variables for nighttime (sleep apnea) and daytime symptoms (daytime napping and oral breathing), showed that the most predictive of snoring were: regular sleep during the day (OR=6.1; 95%CI 1.76-21.04; p<0.005), sleep apnea (OR=4.7; 95%CI 1.63-13.63; p<0.004) and oral breathing (OR=4.5; 95%CI 1.54-12.86; p<0.006).

DISCUSSION

In the first few years of life, the changes in children's sleep patterns are characterized by more sleep consolidation during the night and gradual decrease in daytime naps [13]. The current study, focusing on the habitual sleep times in snoring and nonsnoring 3- to 7-year-old children, demonstrates that the group of children who snore differs from the nonsnoring group in daytime sleep duration, but not in nighttime sleep. For most children, the age of 4 is when the daytime nap begins to disappear from sleep patterns, whereas 47.1% of the children who snore in this study were found to sleep for an average of 52 minutes per day. Longer daytime sleep duration in children who snore may indicate a greater sleep need as compared to nonsnorers. The preschool age is for most children a time when daytime sleep (napping) decreases. In a study by Iglowstein, the most prominent decline in napping habits occurred between 1.5 years of age (about 96.4% of all children take daytime naps) and 4 years of age (35.4%). As shown by reference values for a healthy European (Swiss) population - there are about 8% of children who take naps at the age of 5 years, about 5% at the age of 6 years, about 1% at the age of 7 years [9]. The percentage of children who take naps is dependent not only upon age but also on race factors - e.g. 39.1% of black children at the age of 8 were reported to

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Number of patients 25

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Table I Demographic and	anthronometric dat	ta land sleen characteristics	tor snoring and	I nonsnoring preschool children.
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	Snoring group (n= 34)	Nonsnoring group (n= 66)	p-value
Age (mean, \pm SD, range) ¹	5.38 ± 1.21 3.5 - 7.0	5.67 ±1.12 3,17 - 7,42	NS
Male (%)	15 (44.1)	34 (51.5)	NS
Children from towns (%)	23 (67.6)	38 (57.6)	NS
BMI kg/m ² (mean, ±SD) ² Overweight (BMI percentile >90th)(%) Obesity (BMI percentile >97th) (%)	15.99 ±1.92 4 (11.8) 2 (5.9)	15.88 ±2.32 5 (7.6) 3 (4.5)	NS
	Family history		
Family model (%) parents + 1 child parents + 2 children parents + 3 children and more	16 (47.1) 12 (35.3) 6 (17.6)	22 (33.3) 29 (43.9) 15 (22.7)	NS
Snoring mother (%)	10 (29.4)	8 (12.1)	0.03
Snoring father (%)	21 (61.8)	33 (50.0)	NS
Child sleeps in his/her own bedroom (%)	16 (47.1)	44 (66.7)	NS
Но	ome environment		
Smoking at home (%)	12 (35.3)	19 (28.8)	NS
Pets at home (%)	17 (50.0)	27 (40.9)	NS
Nighttin	ne sleep and symptoms		
Total time of night sleep (mean, ±SD) 7 -8 hours (%) 9 - 10 hours (%) 11 - 12 hours (%)	$10.97 \pm 0.52 \\ 0 (0.0) \\ 24 (70.6) \\ 10 (29.4)$	9.82 ±1.34 7 (10.6)41 (62.1)18 (27.3)	NS
Sleep apnea (%)	24 (70.6)	17 (25.8)	0.001
Bedwetting (%) ³	6 (17.6)	4 (6.1)	NS
Daytim	e sleep and symptoms		
Children with daytime napping (%)3	16 (47.1)	6 (9.1)	0.001
Daytime sleep duration in minutes (mean, ±SD)	51.62 ±28.9	10.70 ± 20.2	0.001
Oral breathing (%)	23 (67.6)	15 (22.7)	0.001
Behavioral problems (%)	14 (41.2)	13 (19.7)	0.02

SD, standard deviation; NS, not significant; Statistical tests used to obtain the p-values:

¹Mann-Whitney U test; ²Student's t-test; ³Yates' chi-square test; the other variable - chi-square test;

nap, compared with only 4.9% of white children. In our study, all participants were Caucasian.

Daytime nap is part of normal individual circadian rhythm. However, when it lasts longer than in peers, may indicate an increased need for rest, especially when accompanied by excessive daytime sleepiness, EDS. In adults, EDS quantified using the Epworth Sleepiness Scale is associated with snoring and OSA. Also in children and adolescent daytime sleepiness is more common in habitual snorers than in nonhabitual snorers as assessed in various questionnaire studies [14,15].

One possible explanation of a longer daytime sleep and daytime sleepiness in children who snore is a greater need for regular sleep during the day due to insufficient nighttime sleep disrupted by respiratory effort–related arousals (e.g. snore arousals), intermittent hypoxia or accompanied oral breathing [1,16]. In a study of O'Brien et al., primary snoring was associated with significant alterations in respiratory arousal (respiratory sleep fragmentation) and reduction of % stadium REM of the total sleep time [3]. In their study, reduced REM%, but not respiratory arousal index, was associated with neurobehavioral dysfunction. Childhood OSA is also predominantly a REM-related disease with involvement of REM-sleep related pharyngeal hypotonia [17,18].

As for a longer daytime sleep, a question arises how nonphysiological breathing route through the mouth and increased work of breathing affect life quality, effort tolerance and need for rest during the day in the study children.

In adults, snoring is mainly associated with excessive daytime sleepiness, whereas in snoring children both EDS and behaviors related to attention-deficit/hyperactivity disorder can be observed [16]. Daytime sleepiness, neurobehavioral problems (in the areas of attention, memory, intelligence, behavioral hyperactivity) and restless sleep are described in habitual snorers [3,19,20]. In the children who snore, Kennedy

Features examined	Odds ratio	-95% CI (lower)	+ 95% CI (upper)	p-value
Age	0.800	0.555	1.153	NS
Gender	1.346	0.580	3.123	NS
BMI	1.024	0.845	1.241	NS
	Family his	tory		
Family model	0.726	0.440	1.198	NS
Family history of snoring	1.528	0.643	3.628	NS
Snoring mother	3.021	1.049	8.697	0.0406
Snoring father	1.615	0.688	3.794	NS
	Personal medic	al history		
Adenotonsillar hypertrophy	4.179	1.688	10.346	0.0023
Recurrent upper airway infections	2.361	0.982	5.673	NS
Gastroesophageal reflux disease	2.370	0.988	5.687	NS
Allergic disease	1.845	0.790	4.307	NS
	Sleep qua	lity		
Total night sleep time	1.523	0.706	3.282	NS
Sleep apnea	6.918	2.722	17.581	0.0001
Oral breathing	7.109	2.799	18.058	0.0001
Bedwetting	3.321	0.854	12.919	NS
	Daytime sym	nptoms		
Excessive daytime sleepiness	4.000	1.577	10.144	0.0039
Daytime sleep	8.889	2.990	26.425	0.0001
Behavioral problems	2.854	1.132	7.196	0.0267

Table 2. Predictive factors of snoring in preschool children.

CI, confidence interval; NS, not significant;

et al. found reduced global IQ, verbal IQ, selective attention, sustained attention and memory index [21]. Different factors, including the genetic ones, are suspected to be involved in the pathogenesis of daytime, neurocognitive consequences of sleep breathing disorders [22]. In our study, the 6-item screening for attention deficits, hyperactivity and impulsivity revealed that behavioral problems were more common in the snoring group. Since we did not compare sleep structure between the groups we can only suspect that behavioral problems in children who snore are related to poor sleep quality.

OSA score was described by Brouillette et al., as a diagnostic test for OSA based on clinical history. In a Carroll et al. study, OSA score had a sensitivity and specificity of 73 and 83%, respectively [10,11]. We used OSA score as a simple, noninvasive method to distinguish snorers from nonsnorers and to find out whether snoring was accompanied by difficult or interrupted breathing. OSA scores between -1 and +3.5 only suggest, though neither confirm nor exclude, the diagnosis of OSA. This group consists of children with varying clinical severity of SDB, mostly in the phase preceding the full clinical OSA pattern, which can be called "pre-OSA". The clinical symptoms registered in "pre-OSA" children indicate obturation of the upper airways (oral breathing, sleep apnea). In practice, only the polysomnographic study allows for

reliable differentiation between primary snoring and snoring concomitant with OSAHS.

Upper airway patency, from nose to larynx, is one of the prerequisites for proper sleep. The degree of upper airway obstruction correlates with many sleep parameters, e.g. the number of times of waking, arousal index and sleep fragmentation [23]. Adenotonsillar hypertrophy (AH) was a risk factor of snoring in our study group, which is not surprising as oropharyngeal lymphoid hypertrophy is a major predisposing factor to SDB in preschool age children [24]. To some extent, AH may explain behavior symptoms in the study children. In research by Suratt et al., snoring in children with lymphoid hypertrophy, alongside shortened nighttime sleep and increased AHI (apnea hypopnea index), was associated with reduced cognitive abilities revealed by tests of vocabulary and similarities [25].

In the study group, no connection was observed between snoring and the amount of nighttime sleep, which in snorers was 11.0 hours on average. In healthy preschool children, Jacklin et al described a total sleep duration to be 11.8 hours at 33 months of age [26]. Gulliford et al reported an average sleep duration of 11.2 hours (11.4 hours) in healthy children at the age of 5 years [27]. In adults, there is a known link between obesity and sleepdisordered breathing (BMI correlates with apnea hypopnea index – AHI) [28]. Also, in infants with body weight of \geq 85th percentile, snoring was most common [29]. In the study group, we found no difference in BMI between the group of snorers and nonsnorers. The relationship between body mass index and sleep-disordered breathing in preschool-age children seems to be weaker, and OSA at this age may be associated with growth delay [30]. Monitoring of anthropometric indices can be useful in predicting the risk of SDB recurrence one year after adenotonsilectomy [31].

Some researchers report on the additional role of other disorders, i.e. gastroesophageal reflux disease and allergic diseases, in the pathomechanism of upper airway obturation [32-34]. These factors may play a part especially in children with ineffective adenotonsilectomy, in the so called residual OSAHS. Generally, it is considered that about 80% of children are cured from SDB by adenotonsilectomy [35]. Allergological studies indicate that snoring risk factors are similar to allergy risk factors, and snoring is included in the spectrum of symptoms of allergic disease in the ARIA guidelines (Allergic Rhinitis and its Impact on Asthma) and in the GINA guidelines (The Global Initiative For Asthma) [36-38]. Atopy is a risk factor for habitual snoring at one year of age [39]. In Chng's research, the strongest predictive snoring factor in children was the coexistence of three atopic diseases: atopic dermatitis, bronchial asthma and allergic rhinitis - OR-7.45 (95% CI, 3.48-15.97) [40]. In this study, screening for atopy in children who snore based on history elicited from parents appeared to be negative but we did not investigate the atopic background by in vitro methods in children who snore.

Marshall et al., in a cohort of five-year-old children with a medical history of rhinitis (participants of CAPS - Childhood Asthma Prevention Study), found a connection between snoring and smoking by the mother in the child's early years of life, first pregnancy birth and the presence of asthma and/or atopic dermatitis at the age of 5 years [37]. They found no link between birth weight, breast-feeding duration, and body mass index (BMI) at 4.5 years of age. In our research, we did not observe more frequent snoring in the first-born children.

The home environment constitutes a major source of exposure to allergens, contaminants and common interior irritants (organic substances, formaldehyde, nitrogen dioxide), which can increase the risk of snoring in children. Forastiere, Liukkonen and others described passive smoking as a factor of snoring in children, while Kalra, just as in our research, found no effect of exposure to environmental tobacco smoke on snoring [39,41,42]. Concluding, in a large population based study the relationship between tobacco home exposure and snoring in children is more distinct.

Snoring in parents increases the risk of snoring in children [14,39]. In our research, snoring occurred more frequently in children of mothers who snore. Genetic predisposition (gene polimorphisms) and environmental risk factors (obesity, OSA, maternal smoking, drinking, medication) could be taken into

consideration to elucidate this relationship, although they were not the main objective of the study [43-45] It should be also considered that many women also start to snore during pregnancy (c. 20%), but we did not have a pregnancy-related question in the questionnaire [46].

There are several potential limitations of the current study. The first is that our data are based on subjective parental reports and not on objective methods (polysomnography, multiple latency sleep test, or actigraphy). It is also unknown whether the group of children who snore with OSA score <+3.5 included children with OSA or with upper airway resistance syndrome (UARS) and were misdiagnosed. Sleep study with the measurement of esophageal pressure (Pes) should be performed as the diagnostic test for UARS. Increased upper airway resistance in this disorder leads to repeated and multiple arousals resulting in an abnormal sleep architecture and daytime sleepiness [47]. The generalizability of findings from this study are limited for one more reason - the children who snore were recruited from those referred to Sleep Laboratory. Thus, a larger study in the population recruited from the local community are required to confirm our findings.

CONCLUSIONS

An increased need for daytime sleep in preschool age children who snore may indicate ineffectiveness of nighttime rest, being indispensable for proper development. In pediatric questionnaire studies, daytime sleep duration is seldom assessed. However, our findings seem to prove the necessity of such an assessment. Pediatric practitioners should pay attention to daytime activity of snoring patients and take into consideration SDB in daytime "sleepers".

In the light of recent research the question arises when we should ignore snoring in preschool children. Guilleminault poses a straightforward question: "Does benign primary snoring ever exist?" [48].

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