CHEST

Original Research

SLEEP DISORDERS

Natural History of Primary Snoring in School-aged Children

A 4-Year Follow-up Study

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Background: **The objective of this study was to examine the natural history of childhood primary snoring (PS) and to identify predictive clinical symptoms and risk factors associated with PS progression to obstructive sleep apnea (OSA).**

Methods: **Children aged 6 to 13 years old who received a diagnosis of PS in our previous community-based OSA prevalence study were invited to undergo repeat polysomnography (PSG) at 4-year follow-up. Subjects with an obstructive apnea hypopnea index (OAHI) ≥ 1 were classified as having OSA at follow-up.**

Results: **Seventy children (60% boys) with a mean age of 14.7 ± 1.8 years were analyzed in this follow-up study. The mean duration of follow-up was 4.6 ± 0.6 years. At follow-up, 26 subjects (37.1%) progressed to OSA, of whom five (7.1%) had moderate to severe disease (OAHI ≥ 5). Twenty-two (31.4%) remained at PS, and 18 (25.7%) had complete resolution of their snoring with normal PSG. Persistent snoring had a positive predictive value of 47.7% and a negative predictive value of 86.4% for progression from PS to OSA. Multivariate logistic regression analysis showed that persistent overweight/obesity was a significant risk factor for the development of OSA at follow-up, with an OR of 7.95 (95% CI, 1.43-44.09).**

Conclusions: **More than one-third of school-aged children with PS progressed to OSA over a 4-year period, although only 7.1% developed moderate to severe disease. Weight control may be an important component in the management of PS because obesity was found to be a significant risk factor for PS progression.** ***CHEST 2013; 143(3):729–735***

Abbreviations: AASM = American Academy of Sleep Medicine; OAHI = obstructive apnea hypopnea index; OSA = obstructive sleep apnea; PS = primary snoring; PSG = polysomnography; SDB = sleep-disordered breathing; Spo_2 nadir = oxygen saturation nadir

Snoring is a common symptom of pediatric sleep-disordered breathing (SDB), and the reported prevalence of habitual snoring ranges from 4.0% to 34.5%.[1-4] SDB includes a spectrum of diseases with severity ranging from primary snoring (PS), to upper airways resistance syndrome, to obstructive sleep apnea (OSA).[5,6] In contrast to OSA, PS, which is defined as snoring without apnea, frequent arousals, or gas exchange abnormalities,[7] has been positioned at the milder end of the SDB severity continuum,[8] and treatment is usually not prescribed.[9]

Nevertheless, whether deferment of treatment of PS is safe has recently led to more research. Kwok et al[10] found that children with PS had increased casual daytime BP and reduced arterial distensibility. Our research group further demonstrated that nighttime BP was also elevated in children with PS.[11] A more

Manuscript received May 14, 2012; revision accepted September 15, 2012.

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Drs Li and Zhu are the joint first authors of this manuscript.

Funding/Support: This study was supported by a Direct Grant for Research from the Chinese University of Hong Kong [Ref. No. 2041773].

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DOI: 10.1378/chest.12-1224

recent study found that PS was a risk factor for hyperactive and inattentive behavior and poor school performance in children.[12] Accumulating evidence suggests PS may be associated with a variety of clinical sequelae, and, therefore, it should no longer be considered as completely benign.[13]

Another important issue that relates to whether PS, if left untreated, progresses to OSA, persists, or resolves over time has been poorly investigated. To our knowledge, only three research studies that examined the natural history of PS in children have been published. The three studies repeated polysomnography (PSG) in cohorts of 20, nine, and 31 children with PS over a 2-year, 3-year, and 6-month period, respectively. All three studies concluded that PS in children generally did not evolve to OSA over time.[14-16] These studies, however, had small sample sizes and consisted of hospital-based subjects. In this study, we aimed to determine (1) the natural history of PS in school-aged children recruited from the community over a 4-year period and (2) the clinical symptoms and risk factors predictive of PS progression to OSA.

Materials and Methods

Subjects

This was a prospective study of a cohort established between 2003 and 2005 for a childhood OSA epidemiologic study.[17] Children aged 6 to 13 years old from 13 primary schools were randomly recruited. A total of 619 subjects underwent PSG, and 161 were defined as having PS (see later discussion in the "Polysomnography" section for definition). For this follow-up study, as a result of limited resources, only the first 99 consecutive subjects with PS were invited to undergo repeat assessment. Subjects were excluded from the study if they had cardiovascular, renal, or neuromuscular diseases; chromosomal abnormalities; or acute illness within 2 weeks of PSG; or if they had undergone upper airway surgery or had started on CPAP treatment during the follow-up period. Written informed consent and assent were obtained from the parents and subjects, respectively. The study was approved by the Clinical Research Ethics Committee of the Chinese University of Hong Kong (CRE_2007.363).

Sleep Symptom Questionnaire

A validated sleep symptom questionnaire[18] was completed by parents of recruited subjects at baseline and follow-up, and the following information was extracted: (1) snoring frequency and other sleep-related symptoms rated on a 6-point scale (0 = never, 1 = less than 1 night per month, 2 = 1 to 2 nights per month, 3 = 1 to 2 nights per week, 4 = 3 nights or more per week, 5 = unclear), snoring and other OSA-related symptoms were defined as present if their frequency scored 2 to 4; (2) clinical features: history of allergic rhinitis and asthma; and (3) socioeconomic and environmental factors. We defined "persistent" as having a positive history at both time points.

Anthropometry Assessment

The weight, height, and Tanner stage of all subjects were assessed on the day of PSG. BMI was calculated as weight/height^2 (kg/m^2). Weight, height, and BMI were converted to z scores appropriate for age and sex, according to local reference.[19] Overweight and obese children were defined as those having a BMI z score ≥ 1.036 and 1.645, corresponding to the 85th and 95th percentile, respectively. We defined "persistent overweight/obesity" as being overweight or obese at both baseline and follow-up. Pubertal stage was evaluated using a self-assessment questionnaire to categorize Tanner stages.[20] Prepubertal was defined as Tanner stage 1, and pubertal was defined as Tanner stage 2 or greater.

Tonsil and Adenoid Size Assessment

The examination was carried out in the morning after overnight PSG by an otorhinolaryngologist. The tonsils and adenoids were evaluated for size by a 4-mm rigid rhinoscope (Storz endoscopy) and a flexible laryngoscope (P4, Olympus), respectively. The sizes of tonsils and adenoids were reported as percentages of the oropharyngeal and nasopharyngeal airways, respectively. A large tonsil or adenoid was defined as the soft tissue occupying $\geq 50\%$ of the corresponding airway. Tonsils and adenoids were further classified as "persistently large" if they were large at both time points.

Polysomnography

All recruited children underwent initial and follow-up standard overnight PSG at a dedicated sleep laboratory with CNS 1000P polygraph (CNS, Inc). In brief, the central and occipital EEG, bilateral electrooculogram, submental electromyogram, bilateral leg electromyogram, and ECG were recorded. The positions of the subject, respiratory airflow (nasal cannula connected to pressure transducer), respiratory efforts (strain gauge), and arterial oxyhemoglobin saturation (by Ohmeda 3700 pulse oximeter) were measured. All data were scored by experienced PSG technologists. At baseline, the standard criteria described in our previous publication were used for scoring,[21,22] whereas at follow-up, the new American Academy of Sleep Medicine (AASM) 2007 pediatric PSG scoring criteria were used.[23] Therefore, all the baseline data of subjects with PS who participated in our follow-up study were rescored using AASM criteria. Those who were not classified as having PS by the new criteria were excluded.

The obstructive apnea hypopnea index (OAHI) was defined as the total number of obstructive apneic and hypopneic episodes per hour of sleep. The oxygen desaturation index was defined as the total number of dips in arterial oxygen saturation $> 3\%$ per hour of sleep. The oxygen saturation nadir (Spo_2 nadir) was also noted. The arousal index was defined as the total number of arousals per hour of sleep.

Children who snored were given a diagnosis of PS if their OAHI was < 1 and Spo_2 nadir was $\geq 90\%$. At follow-up, children were given a diagnosis of OSA if their OAHI was ≥ 1. Normal subjects were defined as nonsnorers with an OAHI < 1 and Spo_2 nadir $\geq 90\%$.

Statistical Analysis

Student t tests, Mann-Whitney U tests, and χ^2 tests for parametric, nonparametric, and categorical data, respectively, were used to detect difference between subjects who participated in this study and those who did not. Paired t tests, Wilcoxon signed rank tests, and McNemar tests for parametric, nonparametric, and categorical data, respectively, were used to examine intragroup differences between baseline and follow-up. Sensitivity, specificity, positive predictive value, negative predictive value, positive likelihood ratio, and negative likelihood ratio, together with their 95% CIs of OSA-related symptoms, were calculated using an online software (http://vassarstats.net/clin1.html). Binary logistic regression analyses were performed separately to investigate the factors