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Original Article

The history of primary snoring in children: the effect of adenotonsillectomy

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ABSTRACT

Primary snoring (PS) is considered as the most benign form of sleep-disordered breathing (SDB), and treatment is usually not prescribed. Studies suggest that PS may not be as benign as had formerly been considered. We aimed to investigate the natural history of PS in children with adenotonsillar hypertrophy, and compare those who underwent adenotonsillectomy (AT) with those who did not.

Material and methods: Children diagnosed with PS based on polysomnographic findings were included in the study. Information retrieved from their medical records, including medical history, physical examination, anthropometric measures, and polysomnography (PSG) results, was reviewed. A telephone interview was conducted 4–6 years following the PSG evaluation. The interview included the Pediatric Sleep Questionnaire Sleep-related Breathing Disorder (PSQ–SRBD) scale, demographics, anthropometric measures, and history of AT.

Results: A total of 248 children (56% males) were studied (mean age: 5.4 ± 3.4 years). Telephone interviews were conducted 5.3 ± 1.1 years following PSG. Sixty-four children (26%) underwent AT/adenoidectomy (A) following PSG. Of the 184 children who did not undergo surgery, 62 (34%) had positive PSQ–SRBD scores five years after diagnosis. Children with PS who underwent AT had better PSQ–SRBD scores at five years post diagnosis than the nonoperated children.

Conclusions: A significant proportion of children with PS persist with SDB symptoms even five years following the diagnosis. In our cohort, a considerable percentage of children with a PSG diagnosis of PS underwent AT despite non-supportive sleep study results. Surgical intervention may have beneficial effects on some children with PS. Further studies using objective measures of sleep and incorporating the effect of SDB duration are required.

1. Introduction

Sleep-disordered breathing (SDB) includes a wide spectrum of disorders with severity ranging from primary snoring (PS) to upper airway resistance syndrome and obstructive sleep apnea (OSA). PS is defined as snoring with no evidence of apnea, gas-exchange abnormalities, or sleep fragmentation as measured during an overnight polysomnography (PSG) evaluation. Its prevalence is about 10% in the pediatric population [1]. PS is considered as the most benign form of SDB, and treatment is usually not prescribed [2].

Nevertheless, recent studies suggest that PS may not be as benign as had formerly been considered [3]. Children with PS reportedly have increased nighttime and daytime blood pressure [4,5]. In addition, PS was found to be a risk factor for hyperactive and inattentive behavior and poor school performance [6]. Increased serum resistin levels were also recently reported in children with PS suggesting increased inflammatory processes [7].

Based on several studies [8–10], the assumption that PS does not progress to more severe forms of SDB is a major reason for not providing treatment to children with PS. A number of studies have recently questioned this assumption [5,11–13]. Li et al. showed that more than one-third of the children with PS progressed to OSA over a 4-year period, and that 7% developed moderate to severe disease [11]. They also found that obesity was a significant risk factor for PS progression.

Despite the current clinical guidelines, some children with PS nevertheless undergo adenotonsillectomy (AT). Data from children with PS who undergo surgical intervention are used to compare between the operated and non-operated children with PS and thus

Abbreviations: PS, Primary Snoring; SDB, Sleep-disordered Breathing; PSG, polysomnography; AT, adenotonsillectomy; A, adenoidectomy; PSQ–SRBD, Pediatric Sleep Questionnaire Sleep-related Breathing Disorder.

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better understand the evolution of the disorder and the related morbidities.

In the present study, we aimed to (1) investigate the natural history of children diagnosed by PSG as having PS over a 4–6-year period, (2) examine the characteristics of children with PS referred for surgical intervention, and (3) investigate the effect of surgical intervention on SDB symptoms in children with PS.

2. Material and methods

The study was approved by the institutional review boards of the Tel Aviv Medical Center and the Ministry of Health (Helsinki Committee), and parental informed consent was obtained. All children referred for suspected SDB due to adenotonsillar hypertrophy to the Pediatric Sleep Center at the Tel Aviv Medical Center between January 2006 and December 2008 and diagnosed as having PS based on PSG were included in the study. All participants underwent a full overnight PSG sleep study (Embla, MedCare diagnostics, Amsterdam, The Netherlands) at the Pediatric Sleep Center.

2.1. Overnight PSG

Chest and abdominal wall movement were monitored by respiratory impedance or inductance plethysmography, and heart rate was monitored by electrocardiography. Air flow was measured by end-tidal capnography, which also provided breath-by-breath assessment of end-tidal carbon dioxide levels ($PETCO_2$; BCI SC-300, Menomonee Falls, WI, USA), as well as by a nasal pressure transducer and an oronasal thermistor. Arterial oxygen saturation (SaO_2) was assessed by pulse oximetry (Nellcor N 100; Nellcor Inc., Hayward, CA, USA), with simultaneous recording of the pulse waveform. Bilateral electrooculogram, eight channels of electroencephalogram, and chin and anterior tibial electromyograms were also monitored. All measures were digitized using a commercially available PSG system (EMBLA, MedCare diagnostics, Amsterdam, The Netherlands). Digital time-synchronized video recording was performed.

Children with snoring throughout the night and apnea–hypopnea index (AHI) <2 per hour of sleep and no evidence for increased work of breathing were considered to have PS [2,14–20]. Subjects with craniofacial anomalies, neuromuscular diseases, metabolic disorders, neurodevelopmental delay, or chronic medical conditions (except asthma/atopy) were excluded. Information retrieved from the children's medical records including medical history, physical examination, anthropometric measures, and PSG results was reviewed. Body mass index (BMI) and the BMI *z*-score at the time of PSG and telephone interview conducted 4–6 years following the PSG evaluation were calculated for each subject. Obesity was defined as a BMI *z*-score of >1.65.

The telephone interview included the Pediatric Sleep Questionnaire Sleep-related Breathing Disorder (PSQ–SRBD) scale, demographics, anthropometric measures (conducted at home by parents) and questions regarding repeated PSG evaluations and ear, nose, and throat (ENT) surgeries (adenoidectomy [A] or AT).

2.2. The pediatric sleep questionnaire sleep-related breathing disorder

The PSQ–SRBD is a well-validated symptom inventory that includes 22 items about snoring, apneas, daytime sleepiness, inattentive/hyperactive behavior, and other SBD features [21]. Responses are "yes" = 1, "no" = 0, or "don't know" (considered missing). The mean response on non-missing items is the total score which can vary from 0 to 1. Higher scores indicate more SDB-related symptoms. A threshold of 0.33, indicating that 33% of the symptom items are positive, is considered a positive screen for pediatric SDB. Subscales within the PSQ–SRBD include a four-item sleepiness scale, a four-item snoring scale, and a six-item inattention and hyperactivity scale derived from the *Diagnostic and Statistical manual of mental Disorders, Fourth Edition criteria for attention-deficit/hyperactivity disorder* [22]. Since its development, this scale has been used in a variety of research settings [21,23]. For the purposes of the current study, we analyzed the total score, as well as the snoring, sleepiness, and the inattentive/hyperactive behavior subscales. In addition, the first two question items concerning habitual snoring ("does your child snore more than half the time?" and "does your child always snore?") were examined separately.

2.3. Data and statistical analysis

The rate of surgical intervention was calculated. In order to study the natural history of PS, we first analyzed the data of children who did not undergo any surgical intervention. The rates of continued snoring and the positive PSQ–SRBD scores were calculated. In order to investigate the effect of surgery on PSQ–SRBD scores and to characterize children who were referred for surgery, we compared the children who underwent surgical intervention with those who did not.

Analyses were performed with SPSS (version 18.0; SPSS Inc. Chicago, IL, USA). Between-group comparisons of continuous variables (age; BMI; AHI; peripheral capillary oxygen saturation, SpO_2; and PSQ–SRBD scores) were conducted with *t*-tests. Dichotomized variables were compared with chi-squared tests. Correlations between variables were performed followed by the calculation of Pearson's correlation coefficients. Linear regressions were used to determine associations after adjusting for potential covariates. All reported *P*-values were two-tailed with statistical significance set at <0.05.

3. Results

A total of 3987 children underwent PSG evaluation for suspected SDB at the Pediatric Sleep Center between 2006 and 2008, of whom 436 were diagnosed as having PS and were candidates for the study. Thirty-eight were excluded due to the underlying congenital anomalies, developmental delay, or other chronic medical conditions. Of the remaining 398 subjects, 26 refused to participate in the study, eight could not respond to the questions due to communication or language difficulties, and 116 could not be located due to incorrect addresses or telephone numbers, leaving a total of 248 children who were studied. Of note, there were no significant differences in age, gender, and BMI *z*-scores at PSG diagnosis between subjects who participated in the study and those who could not be located for the telephone interview. The characteristics and PSG measures at the time of PSG evaluation of the 248 children studied are presented in Table 1.

Telephone interviews were conducted 5.3 ± 1.1 years following PSG. The mean age at the time of those interviews was 10.7 ± 3.7 years, the mean BMI *z*-score 0.09 ± 1.4, and the percentage of obesity 14%.

Sixty-four children (26%) underwent a surgical procedure (A or AT) following the PSG evaluation. Of those, 45 (70%) had A and 19 (30%) had AT. Only one child from the entire cohort had a repeated PSG.

3.1. PSQ–SRBD results in PS with no surgical intervention

To study the natural history of PS, we first analyzed the data of the 184 children who did not undergo any surgical intervention by the time of the follow-up interview. Their results are presented in Table 2. Sixty-five children (34%) had a positive PSQ–SRBD total score.