Obstructive sleep apnea syndrome due to adenotonsillar hypertrophy in infants

Michal Greenfelda, Riva Taumanb, Ari DeRoweb, Yakov Sivana,*

aPediatric Intensive Care, Sackler Faculty of Medicine, Pediatric Pulmonology and Center for Sleep Disorders, Dana Children’s Hospital, Tel Aviv Sourasky Medical Center, Tel Aviv University, 6, Weizman Street, Tel Aviv 64239, Israel
bSackler Faculty of Medicine, Pediatric Otolaryngology Unit, Dana Children’s Hospital, Tel Aviv Sourasky Medical Center, Tel Aviv University, Tel Aviv, Israel

Received 3 January 2003; received in revised form 9 May 2003; accepted 11 May 2003

KEYWORDS
Obstructive sleep apnea syndrome; Infant; Adenotonsillar hypertrophy; Polysomnography

Summary Objective: Adenotonsillar hypertrophy (ATH) is the leading cause for obstructive sleep apnea syndrome (OSAS) in children. The peak age for adenoid and tonsillar hypertrophy and related OSAS is 3–6 years. It has been suggested that OSAS due to ATH is extremely rare in infants. The purpose of the present study was to delineate OSAS due to ATH in infants. Methods: Twenty-nine consecutive infants <18 months of age who underwent polysomnography (PSG) and were diagnosed with OSAS due to ATH were studied. A pediatric sleep questionnaire was completed by parents of all infants. Information regarding demographic variables, referring physician specialty, assessment of infant’s development and recurrence of symptoms post treatment was collected. Pre and post surgery body weight and developmental assessment by parents were evaluated. Results: The mean age of adenotonsillectomy was 12.3 ± 3.9 months with the mean duration of symptoms ranging 6.2 ± 3.0 months. 24% of the infants were born preterm. Snoring was the most common finding and appeared in all infants. Other symptoms were sleep apnea (72%), frequent movements during sleep (69%), mouth breathing (62%) and recurrent awakenings (38%). During the preoperative period, mean body weight decreased from the 67th ± 25 percentile to the 42nd ± 32 percentile (P < 0.00001). 14/29 (48%) of the infants dropped two or more major percentiles before treatment. A significant weight gain to the 59th ± 31 percentile was observed post surgery (P < 0.0001). 5/29 (17%) infants were considered by their parents as having a developmental delay preoperatively, which resolved in 3/5 (60%) post treatment. Clinical symptoms resolved or improved significantly after surgery. Recurrence of symptoms was documented in 6/23 (26%) of infants. Conclusions: Infantile OSAS due to hypertrophic adenoids and tonsils does occur in infants. Unique characteristics for this age group include: male predominance, high incidence of preterm infants, failure to gain weight and high recurrence rate after surgery. Otolaryngologists and pediatricians should be aware to the existence of the “early OSAS” in small infants.

© 2003 Elsevier Ireland Ltd. All rights reserved.
1. Introduction

Adenotonsillar hypertrophy (ATH) is the most common cause of obstructive sleep apnea syndrome (OSAS) in children. OSAS affects about 2% of the pediatric population [1].

The clinical symptoms and polysomnographic characteristics of OSAS in children differ from those in adults. The clinical spectrum of OSAS in children ranges from nocturnal snoring with mild and partial upper airway obstruction to complete cessation of airflow with gas exchange abnormalities and severe disturbance of sleep [2]. OSAS in children may result in severe complications if left untreated. The most important are: growth failure, pulmonary hypertension and neurocognitive deficits such as poor learning, behavioral problems and attention deficit-hyperactive disorders [3,4]. Nevertheless, the diagnosis of OSAS in children is often delayed due to paucity of symptoms and physical signs since the patient is examined in the doctor’s clinic while awake. The gold standard for the diagnosis of OSAS is, therefore, nocturnal polysomnography (PSG), which also assess OSAS severity [5].

The prevalence of OSAS in children peaks at 3–7 years of age parallel to the age of adenotonsillar enlargement. In contrast, OSAS in infancy results usually from congenital facial anomalies and neurologic abnormalities that involve muscle tone and upper airway control. OSAS due to ATH in infants is believed to be very uncommon resulting in relative lower awareness and, therefore, delayed diagnosis. There is a paucity of data on OSAS in infants that is secondary to ATH [6]. This report delineates OSAS due to ATH in infants and highlights its unique features.

2. Patients and methods

The study population consisted of consecutive infants younger than 18 months who were referred by primary community clinics for suspected OSAS. All infants underwent PSG and when the diagnosis of OSAS was confirmed by PSG, each patient underwent further evaluation by the combined pediatric otolaryngologist-pulmonology-sleep team to determine the cause for OSAS. This evaluation included the assessment of ATH by fiberoptic nasopharyngoscopy done by the pediatric otolaryngologists at the same visit to the outpatient clinic. Adenoid hypertrophy was graded as follows:

Grade 1-adenoid obstructing 25% of the choana,
Grade 2-adenoid obstructing 50% of the choana,
Grade 3-adenoid obstructing 75% of the choana,
Grade 4-adenoid obstructing 90–100% of the choana.

Adenoid hypertrophy grade 3 and 4 were considered pathological.

Tonsillar hypertrophy was graded as follows:

Grade 1-tonsils are in the tonsillar pillar,
Grade 2-tonsils are protruding out of the tonsillar pillar,
Grade 3-tonsils reaching midpoint between anterior tonsillar pillar and uvula,
Grade 4-tonsils reach the uvula.

All other infants with OSAS that was not due to ATH and resulted from other causes such as craniofacial anomalies and neurologic abnormalities were not included in the study.

The decision to perform adenotonsillectomy was based on clinical symptoms with OSAS proven by PSG and ATH. In our institution, adenotonsillectomy is the procedure of choice in all infants younger than 2 years of age who are diagnosed with OSAS due to ATH. All infants were otherwise normal healthy infants.

A pediatric sleep questionnaire regarding the infant’s sleep and breathing during sleep was completed by the parents of all infants. This questionnaire was based on OSAS Expanded Childhood Questionnaire [7] and included questions regarding: (a) demographic variables, (b) the referring physician specialty, (c) subjective parental assessment of their infant’s development by milestones achievements and (d) recurrence of symptoms after surgery. Body weight charts of all infants were reviewed from birth and up to 4 years after surgery.

2.1. Polysomnography

All infants underwent overnight PSG for at least 6 h in a quiet, dark room. No sedation or sleep deprivation was used to induce sleep. All children were accompanied by at least one parent throughout the night. During PSG the following variables were continuously measured and recorded by a computerized polysomnograph (Somnostar Alpha Sleep Laboratory, SensorMedics, Yorba Linda, CA, USA):

1) Chest wall and abdominal motions were measured by strain gauges and respiratory inductive plethysmography.
2) Heart rate was measured by electrocardiography (ECG).

3) End-tidal PCO2 (PetCO2) and nasal airflow were measured by capnography (Datex normocap, Datex-Ohmeda Instrumentarium Corp., Helsinki, Finland) sampling at a rate of 60 ml/min.

4) Mouth and nasal airflow was measured by a thermistor positioned at the mouth and nares.

5) Arterial oxygen saturation (SaO2) was recorded by pulse oximetry using a Nellcor, N-200 pulse oximeter set to use 2–3 s averaging (mode 2, fast).

6) Sleep stages were measured using four electroencephalography (EEG) electrodes, located on the scalp, two on each side of the parietal and occipital areas (C3/A2; C4/A1), and by electrooculography (EOG).

7) Submental electromyography (EMG) was measured by two electrodes located at the point of chin and belly of the digastric muscle on each side of the chin.

8) Children were also monitored and recorded on audio–videotape using an infrared video camera.

Each child was continuously observed by a technician trained in pediatric PSG who also recorded children’s sleep behavior and respiratory events.

The following variables were measured and analyzed.

2.2. Respiratory variables

The analysis of the PSG was based on accepted guidelines [8–10]. In brief, obstructive apnea was defined as cessation of airflow at the nose and the mouth, as measured by the thermistor and the capnograph, while the respiratory effort continues (movements of the rib cage and the abdomen). The number and duration of obstructive apneas of any length were quantitated.

Obstructive hypopnea was defined as a 50% or greater decrease in the amplitude of the nasal/oral airflow signal, as measured by the thermistor, accompanied with paradoxical chest and abdominal movement, and usually by hypoxemia or arousal.

Apnea and hypopnea indexes were calculated by dividing the number of apneas or hypopneas by the hours of sleep.

All hemoglobin desaturations defined as decreases > 4% from baseline SaO2 or < 92% were quantitated. Measurements associated with poor pulse tracings were excluded.

PetCO2 abnormalities were recorded and quantitated according to Marcus et al. [10].

2.3. Sleep architecture

Sleep was staged according to standard criteria [11] using EEG, EOG and EMG. Sleep onset was defined as two consecutive 30-s epochs of Stage I or one epoch of Stage II or rapid eye movement (REM) sleep. Total sleep time (TST) was defined as all sleep incurred from sleep onset until morning awakening. Sleep efficiency was defined as the sleep time minus the time of wakefulness out of TST. Respiratory parameters were expressed as a percentage of the TST.

2.4. Statistical analysis

Descriptive statistical parameters include presentation of means and standard deviations. Statistical analysis was performed using BMDP Statistical Software (1993), chief editor: W.J. Dixon, University of California Press, LA.

Analysis of variance (ANOVA), ANOVA with restricted measures and Pearson’s \( \chi^2 \)-test or Fisher’s exact test were used as appropriate. \( P \) values of < 0.05 was considered significant.

3. Results

The study population consisted of 29 infants (20 boys and nine girls). OSAS was diagnosed at a mean age of 12.3 ± 3.9 months and was followed by adenotonsillectomy within 2 weeks in 27/29 infants. Two infants underwent adenoidectomy alone because tonsils were not enlarged (grade 1).

Infant’s characteristics are presented in Table 1. All infants snored. The presenting symptoms are shown in Fig. 1. The specialty and sub-specialty of the referring physicians were: general otolaryngologists (55%), general pediatricians (24%), pediatric otolaryngologists (17%) and family physicians (4%). Symptoms of OSAS started at a mean age of 6.1 ± 3.2 (5–17). The overall mean time interval be-
between the beginning of symptoms and diagnosis was 6.2 ± 3.0 months (2–12). Infants who were referred by pediatric otolaryngologists had shorter mean symptoms duration prior to diagnosis (4.8 months) compared with general otolaryngologists (6.5 months), general pediatricians (6.4 months) and family physicians (7 months) (P = NS). During the preoperative period, mean body weight decreased from the 67th ± 25 percentile in the asymptomatic period before the symptoms of OSAS started, to the 42nd ± 32 percentile immediately prior to surgery (P < 0.00001). A significant weight gain to the 59th ± 31 percentile was observed post surgery (P < 0.0001).

No significant differences were found between body weight percentiles during the asymptomatic preoperative period to the post surgery period. In 14/29 (48%) infants a drop of two or more major percentiles was observed before treatment. This sub-group did not have any unique characteristics compared with the other infants in the study and did not differ in age, gender, symptoms duration and the referring physician specialty. Based on milestones achievements, 5/29 (17%) of infants were considered by their parents as having developmental delay, which resolved in 3/5 infants after surgery.

Clinical symptoms resolved or improved significantly after surgery. Snoring disappeared in all cases. However, 6/23 (26%) of infants had recurrence of symptoms during the follow-up period. The earliest postoperative interval to recurrence of symptoms was 6 months. After repeat PSG and further evaluation including a repeat fiberoptic nasopharyngoscopy, they underwent recurrent adenoidectomy. All recurrences and surgical revisions were done in infants who previously underwent both adenoidectomy and tonsillectomy. The recurrent procedure took place at least 18 months after the first. This subgroup did not have any unique characteristics regarding age, gender and history of prematurity.

4. Discussion

OSAS due to ATH in the pediatric population is relatively common especially in young children 3 years and older. Its clinical characteristics are well defined. In contrast, OSAS secondary to ATH is considered very uncommon in infants and only a paucity of information is found in the literature [3,6,12].

The hypothesis that OSAS in children without neuromuscular disease is a structural abnormality has been questioned in recent years based on several observations: patients with OSAS do not obstruct during wakefulness, no correlation exists between adenotonsillar size and OSAS, a small percentage of children are not cured by adenoten-
sillectomy and 98% of children have no sleep-disordered breathing even though ATH is common in this age group [13].

OSAS is now thought to be caused by a dynamic process resulting from a contribution of structural upper airway narrowing and abnormal upper airway neuromotor tone. The combination of both these mechanisms is responsible for the increased upper airway collapsibility during sleep [13]. Activation of the upper airway muscles and increasing their tone compensates for structural narrowing. This compensatory mechanism may be less efficient during sleep.

It has been suggested that children with OSAS have a tendency for upper airway collapsibility due to centrally mediated abnormalities in the activation of upper airway muscles [14]. An elevated arousal threshold in response to hypercapnia was also observed in children with OSAS [15].

While ATH is the principal structural component in older children, congenital syndromes and malformations account for the vast majority of anatomical narrowing in infantile OSAS. The leading anomalies are micrognatia either as an isolated anomaly or as a part of abnormalities such as the Pierre Robin sequence and Treacher Collins syndrome [16,17]. Mid-face hypoplasia occurs in achondroplasia, Crouzon syndrome and Apert syndrome [18,19], and macroglossia is common in Down’s syndrome and in infants with mucopolysaccharidoses [20,21]. In these abnormalities the upper airways are narrowed and predispose the infant to OSAS.

Disorders where decreased upper airway muscle tone is the major component in the pathogenesis of OSAS include neuromuscular disorders such as Werdnig–Hoffmann and muscular dystrophies.

The present study describes OSAS due to ATH in infants younger than 18 months of age. The data show that OSAS in this age group share a few common characteristics with OSAS due to ATH in older children. Snoring occurred in all infants and sleep apnea, body movements, mouth breathing and awakenings were also observed, although the incidence of the latter was relatively low [7,22].

Nevertheless, some characteristics may be distinctive to infants and are uncommon in older children.

A male preponderance (2:1) was observed in this report compared with the equal gender distribution of the syndrome in preschool and older children [23].

One quarter of the patients in this series were born preterm. The reason for this high rate and the occurrence of OSAS in infants who were born preterm is worth further investigation. Possible explanations may include the higher incidence of muscle hypo-tonicity, which is more frequent in infants who were born preterm, and chronic inflammation stimulating local tissue growth in the nasal airways due to prolonged intubation periods.

Failure to thrive (FTT) was a common finding in this series. The association of OSAS to FTT has been well documented in older children [3]. Due to greater awareness, overt FTT is infrequent now and occurs only late in untreated OSAS cases [7]. Possible mechanisms for poor weight gain include anorexia and dysphagia resulting from the ATH [6], abnormal secretion of growth hormone and insulin growth factor-1 [24,25] and increased caloric expenditure due to increased work of breathing during sleep [25]. Although the overall rate of FTT due to OSAS secondary to ATH in the entire infant population may be low because OSAS in this age group is infrequent, it should, nevertheless, be considered in the differential diagnosis and the evaluation of infants with FTT. Most infants in this report demonstrated a catch-up growth after adenotonsillectomy. This has also been showed by others [26].

Post surgical adenoid tissue regrowth and recurrence of previously resolved OSAS is seldom reported [27]. In our series, one quarter of the infants demonstrated symptom recurrence, and after confirmation of OSAS by PSG they required repeated adenoidectomy. The chances of incomplete excision during the first operation may be higher in infants due to technical difficulties, small airway dimensions and unidentified residual adenoid tissue. Other additional explanations include the co-existence of anatomical and neurological factors such as narrow oropharynx and hypotony of the pharyngeal muscles. GER could also be a predisposing factor for adenoidal regrowth.

A trend towards higher awareness of pediatric otolaryngologists to the possibility of OSAS due to ATH in infants was observed. The difference did not reach statistical significance probably due to the small sample size. This may result from the higher expertise and experience of physicians with subspecialty training.

In conclusion, “infantile OSAS” as caused by ATH, may be defined as a unique entity occurring in young infants that share common features with “classical” OSAS of childhood, but is distinctive by male predominance, high incidence of prematurity, significant FTT and a relatively high recurrence rate. Physicians caring for infants should be aware of the possibility of OSAS and its unique characteristics in this age group.
Acknowledgements

The authors thank Pnina Lilos from the statistics laboratory of the Tel-Aviv University for the statistical analysis.

References


