OBSTRUCTIVE SLEEP APNEA SYNDROME IN THAI CHILDREN DIAGNOSED BY POLYSOMNOGRAPHY

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Abstract. Overnight polysomnography was conducted in 39 Thai children with clinically suspected obstructive sleep apnea syndrome (OSAS) during the years 1994 to 1996. Eighty-five percent of these children met the polysomnographic criteria of pediatric OSAS, 42.4% among whom had severe OSAS. Male : female ratio of children with OSAS was 4.5:1. The peak age at the time of diagnosis was 3 to 4 years. The most common predisposing factor was adenoidal and tonsillar hypertrophy. Adenoidectomy and/or tonsillectomy was the most effective therapeutic option. Recovery of symptoms was observed following surgery and nasal continuous positive airway pressure.

INTRODUCTION

Obstructive sleep apnea syndrome (OSAS) is a syndrome of disordered breathing characterized by a combination of intermittent partial and/or complete airway obstruction occurring during sleep leading to hypoventilation (hypoxemia and hypercapnia) and sleep disturbance (Loughlin, 1992). If left untreated, it can result in significant morbidity in children. It has been shown to result in neurodevelopmental problems such as excessive daytime somnolence, behavioral disturbances, school failure, failure to thrive and developmental delay (Brouilette *et al*, 1982). Chronic hypoxemia and hypoventilation can lead to pulmonary hypertension, congestive heart failure (Brouilette *et al*, 1982), and even death (Bradley and Phillipson, 1985).

Although precise incidence figures are not available, recent study suggests that the prevalence of OSAS in 4- to 5-year-old children is approximately 0.7% (Ali *et al*, 1993). Diagnosis of childhood OSAS by history alone or any combination of history plus physical examination or laboratory tests while awake is not certainly reliable (Carroll *et al*, 1995). Overnight polysomnography (PSG) has remained the "gold standard" for establishing the diagnosis and determination of the severity of childhood OSAS (American Thoracic Society, 1996; Carroll and Loughlin, 1995).

In Thailand, although pediatricians and otolaryngologists may see a lot of patients with clinically suspected OSAS, without polysomnography the diagnosis of OSAS cannot be definitely established. Therefore the actual incidence, clinical and polysomnographic manifestations of OSAS in Thai children have not yet been reported. As there has been an increasing awareness of OSAS and its complications, we recently set up a pediatric sleep laboratory using polysomnography at Ramathibodi Hospital, Bangkok in the year 1994. The objectives of the present study were to investigate the incidence of OSAS diagnosed by overnight polysomnography among Thai children with clinically suspected OSAS and to demonstrate clinical and polysomnographic findings of OSAS in Thai children including clinical histories and predisposing factors.

PATIENTS AND METHODS

Children, aged under 15 years old, referred to pediatric chest clinic at Ramathibodi Hospital for evaluation of OSAS, were sequentially recruited from August 1994 to March 1996. All children had a history suggesting partial or complete upper airway obstruction during sleep. For each child, evaluation included complete history, physical examination and radiograph of the upper airway. Clinical histories obtained from parents were supplemented by childhood OSAS questionnaires (Carroll *et al*, 1995). Overnight polysomnography (PSG), (Poly G®) was performed on every child, including oronasal airflow (thermister), chest and abdominal pneumobelts, pulse oximetry and electrocardiogram (EKG). The child's sleep behavior and respiratory pattern were also continuously observed and recorded by a special nurse trained in polysomnography.

The PSG respiratory events were visually reviewed and manually edited. Obstructive apnea was defined as the absence of oronasal airflow in the presence of chest wall motion. The number and duration of obstructive apnea lasting for two or more consecutive breaths (Carroll and Loughlin, 1992) were determined. The obstructive apnea index, defined as the number of obstructive apneas per hour of sleep, was calculated. Central apnea was defined as the absence of chest wall motion associated with absence of oronasal airflow detected by thermister. We defined the lowest oxygen saturation as that measured by the pulse oximetry while the patient was asleep without any movement artifact and where the pulse rate detected by the oximeter was comparable to the heart rate detected by EKG. The presence of paradoxical inward rib cage motion on inspiration indicating airway obstruction was determined by the display of PSG tracings corresponding with the nurse's observation.

The child was considered to have obstructive sleep apnea (OSA) if the lowest SpO_2 was lower than 92% and the obstructive apnea index (OAI) was greater than one per hour of sleep time (Marcus *et al*, 1992) or if the desaturation episodes (< 92%) were associated with the presence of obstructive apnea and/or paradoxical inward rib cage motion on inspiration. Since there was no consensus on guidelines for rating the severity of PSG findings, we therefore defined the severity of the patients who had nadir $SpO_2 \leq 75\%$ and/or OAI ≥ 10 per hour of sleep time as severe OSA. OSA which did not meet the criteria for severe OSA was defined as mild to moderate OSA. Primary snoring was characterized by the presence of snoring during sleep without associated apnea or desaturation.

Data are expressed as percent or mean ± standard deviation where appropriate.

RESULTS

Clinical data

Of 39 children with clinically suspected OSA, 33 (84.6%) had OSAS, 3 (7.7%) had primary snoring, 2 (5.1%) had normal PSG study and 1 (2.6%) had central apnea associated with desaturation.

A total of 33 children who met the PSG criteria for OSAS were analyzed in detail. Ages of children with OSAS ranged from 3 months to 12 years, with a mean age of 5.1 ± 3.1 years. The age distribution is shown in Fig 1; the peak age of affected children was between 3 and 4 years. Twenty-seven (81.8%) were male. According to the Thai standard growth curve, 16 (48.5%) children had normal weight, 10 (30.3%) were obese (weight greater than 120% of ideal body weight by height) and 7 (21.2%) were underweight (weight less than 90% of 50th percentile of body weight by age).

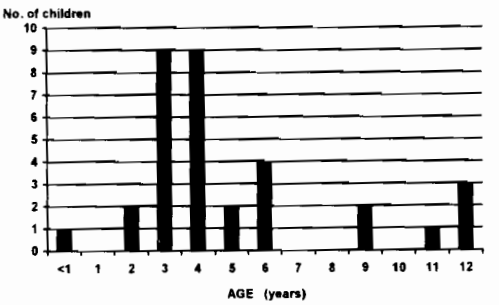

Fig 1–The number of Thai children with OSAS at each year of age.

Clinical histories obtained using the childhood OSAS questionnaires are shown in Table 1. There were 3 children whose questionnaires were not completed. The most common parental observations were the child's struggling to breathe (87%), snoring loudly or extremely loudly (77%), and snoring "most nights" (70%). Only 34% and 18% of all parents were able to observe obstructive apnea and cyanosis during sleep respectively. Hypersomnolence which the parents described as "the child often fell asleep in class" was reported only in one of 19 school aged children. Among 18 children who were older than 4 years old, nocturnal enuresis (most nights) was present in 4 (22%). The oldest child presented with enuresis was an 11 year old girl with Crouzon's syndrome, a known genetic defect. Hyperactivity was noted by parents of 2 children; one child was diagnosed by a primary pediatrician as having an anxiety disorder. Diazepam was once prescribed to this child in error due to hyperactive behavior during the daytime and frequent awakenings during sleep.

Hypertrophied tonsils or adenoids were the most frequent predisposing factors of OSA; this occurred in 26 children (78.8%). Other predisposing factors

Table 1

Historical features of Thai children with OSAS*.

	%
Parent's observations during sleep	
- Observed child struggling to breathe (28)	86
- Watches child sleeping, afraid about breathing (29)	66
- Shake child to make him or her breathe (29)	45
- Observed obstructive apnea (29)	34
- Observed cyanosis (28)	18
- Observed excessive movement (29)	17
Snoring	
- Snoring loud or extremely loud (30)	77
- Snoring "most nights" (30)	70
Other symptoms	
- mouth breathing "most nights" (30)	53
- upper respiratory tract infection > 6 times per year (29)	45
- nasal voice (27)	44
- nocturnal enuresis "most nights" in > 4 year old children (18)	22
- mouth breathing "most days" (30)	13
- hyperactive (30)	7
Weight	
- Obese (33)	30
- Underweight (33)	21

* Number of children whose parents responded to that question are in parenthesis.

are obesity (30.3%), Crouzon's syndrome (3%) and micronagthia with hypotonia (3%). There were 5 children whose predisposing factors were both hypertrophied tonsils/adenoids and obesity.

Polysomnographic features

The average total sleep time was 7.3 ± 0.9 hours. The lowest SpO_2 was in the range 50-91%; the mean average lowest SpO_2 was76 ± 11.8%. The maximum OAI was 19 per hour of sleep time. Fourteen (42.4%) children met the criteria for severe OSA (12 children had lowest $SpO_2 \leq 75\%$; 2 children had lowest SpO_2 of 76 and 79% but their OAI were 12 and 18 per hour of sleep time respectively).

Treatment and outcome

Tonsillectomy or adenoidectomy or both were performed in 15 children (45.5%) who had severe OSAS associated with tonsil and adenoid hypertrophy. The youngest child (3 months old) with micrognathia and generalized hypotonia diagnosed polysomnographically as severe OSA required tracheostomy. No surgical complications were observed. All children improved after surgical relief of airway obstruction, having less labored breathing during sleep, quieter and more restful sleep. Of 4 underweight children who had surgical treatment, 3 had normalization of growth curve after the surgery. We were able to repeat polysomnographic study in one child with severe OSA seven days following the surgery. The lowest SpO_2 went up from 59% to 88% and OAI returned to normal range. Paradoxical inward rib cage motion was also significantly reduced and snoring disappeared.

Non-surgical management included dietary control in all 10 obese children, pharmacologic treatment including antihistamines, decongestants, ketotifen and/or nasal steroids in 10 children with mild to moderate OSA and mask positive airway pres-

sure in 2 children with severe OSA.

Apart from dietary control, 5 of 10 obese children underwent tonsillectomy and adenoidectomy due to concomitant presence of tonsillar and adenoidal hypertrophy. One obese child required Bilevel positive airway pressure (BiPAP®) during sleep. Clinical improvement was observed in all 5 obese children who had surgery whereas the outcome was variable among the group of obese children on whom the surgery was not performed. Weight reduction was successful only in 4 of 10 obese children. BiPAP® which was one of mask positive airway pressure device was instituted in a 12-year-old obese boy with severe OSA whose tonsils and adenoids were not enlarged. Because he had such a poor weight control and OSA symptoms persisted and tended to be worse, we strongly recommended BiPAP ventilator for him during sleep. He spent almost two months getting accustomed to a nasal mask and BiPAP ventilator. We repeated three nights of PSG studies while he was put on different pressure levels of BiPAP. On the optimum ventilatory setting with inspiratory pressure of 8 and expiratory pressure of 2 cmH_2O, he had a remarkable improvement in PSG findings; lowest SpO_2 was increased from 76% to 92% and OAI was decreased from 12 to 0.83 per sleep hour. Moreover, no paradoxical movement of chest and abdomen or snoring were observed. His school performance was clearly improved thereafter.

An 11-year-old girl with Crouzon's syndrome who had severe OSA was treated with nasal continuous positive airway pressure (CPAP). She also spent almost 6 weeks to get used to the support ventilatory system. We repeated PSG while she was on CPAP 5 cmH_2O. Her PSG findings were considerably improved; lowest SpO_2 went up from 70% to 88% and OAI was decreased from 12 to 0.9 per sleep hour. In addition her enuresis was ceased after she had been on CPAP during sleep. No side effect of nasal CPAP other than nasal congestion was observed.

DISCUSSION

We found a high incidence of OSAS among Thai children who were referred to our chest clinic for a history of breathing difficulty during sleep. Eighty-five percent of patients studied met the polysomnographic criteria of OSAS, 42% among whom were found to have nadir $SpO_2 < 75\%$ and/or OAI > 10 per hour of sleep time which we classified as severe OSAS. The incidence of OSAS in this study was much higher than that presented in another recent study (Leach *et al*, 1992) which found that only 37% of children referred actually had OSAS. This could probably be explained by the fact that OSAS has received relatively little attention by Thai physicians. Children with OSAS might be referred for evaluation later in the course of the disease. That was also a reason why we found a high percentage of severe OSAS. In addition, the polysomnographic criteria of OSAS used in our study also differed from the study of Leach *et al* (1992) in which the number of OAI was not measured and the level of lowest oxygen saturation by which OSAS was determined was not specified. This may have resulted in the recording of a higher incidence of OSAS among our patients. In our institution, since the incidence of OSAS was high among patients with clinically suspected OSAS, overnight polysomnography would therefore be recommended in most cases. Polysomnography assists us in differentiating OSAS from primary snoring and central apnea. It quantifies the severity of OSAS, guides the treatment, anticipates the risk of post operative respiratory compromise and provides a baseline for the follow-up.

According to our findings, OSAS appeared to be more prevalent in boys than in girls. The male: female ratio was 4.5:1. Our sex ratio differed from that reported in western countries which found that the sex ratio for OSAS in prepubertal children was roughly 1:1 (Guilleminault *et al*, 1981; Frank *et al*, 1993). In adults, OSAS occurs more commonly in males than in females (Young *et al*, 1983). The administration of exogenous testosterone may result in OSAS (Schneider *et al*, 1986). These findings suggest the possibility that androgens play a role in pathophysiology of OSAS, and that female sex hormones may play a protective role. The difference in sex ratios between Thai and Caucasian children requires further investigation in this context.

Our data indicate that the peak age of childhood OSAS at the time of diagnosis was 3-4 years, similar to a previous study in USA (Brouillette *et al*, 1982). This age group may be particularly susceptible because of the prominence of pharyngeal lymphoid tissue (Jeans *et al*, 1981) and probably frequent upper respiratory infections. We found

that in Thai children tonsillar and adenoidal hypertrophy were the most common predisposing factors, which was similar to the findings previously reported (Brouillette *et al*, 1982; Frank *et al*, 1983).

A majority of parents' observations during sleep were "snoring" and "observed child struggling to breathe". Only a minority of parents were able to observe "cyanosis" and "obstructive apnea". Although excessive daytime sleepiness is a cardinal symptom of sleep apnea in adults, occurring in 78% (Guilleminault *et al*, 1978), hypersomnolence in this study which the parents described as "the child often fell asleep in class" was reported only in one of 19 school-aged children. Other studies in children similarly found lower incidence of hypersomnolence in children than in adults (Frank *et al*, 1983; Richardson *et al*, 1980). Of the cases noted, hyperactivity was observed in two children, probably due to an attempt to keep them from falling asleep during the day (Mandel and Reynold, 1981). Enuresis caused by OSAS occurred in a girl with Crouzon's syndrome. She became dry after correction of OSAS with nasal CPAP. It was speculated that the improvement of enuresis was due to elimination of sleep disruption and the subsequent normalization of arousal mechanisms (Weider *et al*, 1991).

Although the PSG information obtained from our polysomnographic device was not complete as recommended by the American Thoracic Society (American Thoracic Society, 1996), we were able to document the essential diagnostic elements of OSAS, *ie* obstructive apnea, central apnea, oxygen desaturation, chest and abdominal movement, body position, tachycardia and bradycardia. The PolyG® polysomnographic device used in this study has been recently assessed for accuracy by comparing the polysomnographic findings of PolyG® with a gold standard polysomnography. The results indicated that the overall accuracy of this device was greater than 90% (Man, 1994). In our study we trained a team of special pediatric nurses who could provide us additional information of OSAS including sleep *vs* wakefulness, the loudness of snoring, labored breathing, sweating and presence of paradoxical inspiratory rib-cage movement while the patient was asleep. The information obtained from both the polysomnographic device and records by attending special nurses would thus be adequate to diagnose OSAS.

Our PSG criteria used to diagnose OSAS have followed the most recent consensus recommendations of the American Thoracic Society (American Thoracic Society, 1996) which stated that the number of obstructive apnea exceeding 1 apnea per hour and the presence of partial airway obstruction associated with paradoxical inspiratory rib-cage movements should be considered abnormal. The level of nadir oxygen saturation was considered abnormal if SpO_2 was less than 92%. An arbitrary cut-off level of abnormal oxygen saturation was recommended by Marcus *et al* (1992) who showed that the lowest mean SpO_2 in normal sleeping children was 96 ± 2%. On this basis SpO_2 values < 92% have been considered abnormal in the pediatric age group. Due to the unavailability of an end tidal CO_2 monitoring device, we did not include the end tidal CO_2 level for the criteria of OSAS and the severity of hypoventilation could not be quantified.

As in previous series (Suen *et al*, 1995), tonsillectomy and/or adenoidectomy were the most effective measures in relieving the OSAS. All patients had improvement in respiratory disturbances after surgery. There were no immediate and long term post-operative complications. Three of four underweight children showed improved growth after surgery. Their energy expenditure during sleep might decrease after recovery of OSAS. The poor growth seen in some children with OSAS may be a consequence of increased caloric expenditure caused by increased work of breathing during sleep (Marcus *et al*, 1994).

One girl with Crouzon's syndrome and one boy with obesity but without adenoidal and tonsillar hypertrophy were treated with nasal CPAP and nasal bi-level positive airway pressure (BiPAP®) respectively. The positive airway pressure created by the equipment can be thought of as "pneumatic splinting" of the collapsible segment of the upper airway. Although a prolonged period of time was required to accustom both patients to the positive airway pressure system, CPAP and BiPAP were effective and well-tolerated. We selected appropriate pressure by titration of pressure while performing additional overnight polysomnography. The only complication was chronic nasal congestion seen in the girl with Crouzon's syndrome. Since the main limitation of successful CPAP use appears to be poor compliance (Marcus *et al*, 1995), the importance of using CPAP and BiPAP during sleep was emphasized periodically to patients and their parents. The obese boy reported better school

performance after using BiPAP. Several studies have shown correlation between nocturnal hypoxemia and daytime cognitive impairment (Greenberg *et al*, 1987; Findley *et al*, 1986). It is likely that hypoxemia occurring in this child during sleep impaired his cognitive function during the daytime. The correction of oxygen desaturation consequently improved his school performance.

For all obese patients with OSAS, weight reduction was strongly recommended, but it was difficult to achieve (Mallory and Beckerman, 1992). In this study, weight reduction was successful only in less than one third of obese children. Improvement of symptoms was noted in all five obese children who underwent adenotonsillectomy. In Thailand, unlike surgery, the expense of CPAP or BiPAP is not covered by the government health service or third party insurance and it is more expensive than performing adenotonsillectomy. Accordingly adenotonsillectomy may be indicated as a first palliative measure in obese children, particularly those with chronic adenoidal and tonsillar hypertrophy.

In summary, we found that Thai children with a history of breathing difficulty during sleep demonstrate a high incidence of OSAS. Thai boys are more prone to OSAS than girls. The peak age at the time of diagnosis is 3-4 years old. The most common predisposing factor is adenotonsillar hypertrophy. An overnight polysomnography is a valuable tool for the diagnosis and assessment of the severity of OSAS. Adenoidectomy and tonsillectomy are the most cost effective therapeutic options in pediatric OSAS.

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