Clinical and Therapeutic Aspects of Obstructive Sleep Apnea Syndrome in Infants and Children

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Summary: Sleep related increases in respiratory resistive loads in infants and children may or may not be associated with complete obstruction, i.e. obstructive apneas. Nevertheless, important clinical symptoms, both nocturnal and diurnal, may be present and should be taken into consideration. A thorough investigation must be performed to select the most appropriate therapy.

Obstructive sleep apnea syndrome (OSAS) was first identified by Guilleminault et al. in 1976 (1). A few years later, the same group emphasized that the clinical diurnal and nocturnal symptoms of this syndrome were not always associated with complete obstruction of the upper airway during sleep (2). Sleep-related increases in respiratory resistive loads are associated with significant or important clinical symptoms and should be considered in the search for optimal therapeutic strategies. Therefore, we briefly review the clinical and therapeutic aspects of sleep-related respiratory resistive loads, both with and without associated obstructive apneas, in infants and children.

FREQUENCY

We still know little about the prevalence of increased respiratory resistive loads during sleep in infants and children. The frequency of heavy snoring in the age range 4–6 years was reported to be 7.1 to 9.7% (3,4). In one of these studies, children were studied at home with oximetry and video examinations. The estimated percent of children with OSAS was 1.75–2.2%. Another study, reporting a wider age range (6 months to 6 years), estimated the frequency of OSAS at 1.6–3.4% on the basis of a questionnaire (5).

CLINICAL SYMPTOMS

Clinical symptoms associated with sleep-related respiratory resistive loads in infants and children are both nocturnal and diurnal. Several reports have concerned the frequency of these symptoms (2,6–10). Only two, however, have compared this frequency between a study group and the general pediatric population (9,10). Nocturnal symptoms are heavy snoring, difficulty breathing, respiratory pauses, restless sleep and abnormal movements, profuse nocturnal sweating, special sleeping position and enuresis that can be assessed only during childhood. Diurnal symptoms include excessive daytime somnolence, which is difficult to diagnose in infants and young children, disturbed sleeping-waking rhythms, mouth breathing and, in older children, abnormal behavior, including hyperreactivity, aggressive and rebellious behavior, learning problems and morning headaches. Upper airway infections are frequent in younger children (7). Failure to thrive occurs in one-third to one-half of those with sleep-related upper airway abnormalities and can delay diagnosis and treatment. Obesity is rare.

Cardiac failure associated with systemic hypertension may be the presenting problem and may be associated with acute upper airway infection (11). In most reported cases, clinical symptoms were present, but ignored. Because cardiovascular symptoms are frequent in adults with OSA, it is of interest to review their frequency in infants and children without cardiac failure. However, few data are available (7,12,13). In a series of 22 cases, right ventricular hypertrophy was reported in 55% of the cases on X-ray and/or echocardiography examination (7). Interestingly, it has been shown that right ventricular ejection fraction significantly increases after adenotonsillectomy (13). Recent advances in echocardiography allow examination of interventricular septum behavior during sleep-related
upper airway obstruction. In one 11-year-old child, Guilleminault et al. reported a leftward shift of the septum, even during heavy snoring (14). Cardiac ECG abnormalities have been examined in a group of 50 infants and children with OSAS (6). Sinus arrests lasting from 2.5 to 9 seconds were present in 52% of the cases, second-degree atrio-ventricular block in 28% and paroxysmal tachycardia in 16%.

In a series of 50 cases, diurnal systemic hypertension was found in 10% of the cases, all older than 10 years (6). More recently, no abnormalities in systolic or diastolic pressure were observed in 22 children aged 6 (± 0.4) years (15). Noninvasive continuous blood pressure measurements have been performed in two children whose fingers were large enough to wear the finger cuff of the equipment, one of whom manifested pulsus paradoxus (14).

Finally, there have been reports of sudden death in children with OSAS (16). The percentage of upper airway abnormalities is variable among reports of large groups of infants and children with OSAS. The variability may be explained by the different age ranges of the groups studied and the different clinical orientation of the sleep laboratories (6–8). In most reports, hypertrophy of the tonsils and adenoids is the major cause of OSAS. Facial dysmorphia was the cause of OSAS in 32% of the cases in one report (6). This includes micrognathia-retrogionathy, Pierre Robin syndrome, Crouzon’s disease and Down’s syndrome. Sleep-related disordered breathing due to abnormalities of the upper airway is observed in neuromuscular disorders including Arnold-Chiari malformation, syringomyelo-bulbia and myotonic dystrophy. General disorders such as congenital myxedema, Prader Willi syndrome, obesity and sickle cell anemia may be involved (17). Finally, laryngomalacia can be responsible for OSAS.

**DIAGNOSIS**

The gold standard for diagnosis of sleep-related increased resistive loads with or without obstructive apneas is a nocturnal polysomnographic recording. However, some groups have proposed that the diagnosis may be established on the basis of a standardized questionnaire (9) with or without oximetry and video examination at home (10). From a practical point of view, afternoon naps are useful in infants and children younger than 4 years old. However, for afternoon naps to be helpful for diagnosis, they must include a period of rapid eye movement (REM) sleep. Afternoon naps without an REM sleep period should be followed by a nocturnal sleep study. In all sleep studies, neither sleep deprivation (18) nor chloral hydrate (19) should be used.

Polysomnographic findings can be divided into two categories, i.e. with or without obstructive apneas. Obstructive apneas are not always present, despite clinical symptoms of sleep-related disorders. Furthermore, there are few normative data on obstructive apnea occurrence in a normal population of infants and children. They are available for infants younger than 6 months (20) and for children (21).

When polysomnographic data include obstructive apneas, they are usually longer and more frequent during REM sleep. Obstructive apneas are associated with a fall in arterial oxygen saturation (SaO2). No behavioral arousal was observed in a group of prepubertal children with OSAS, although arousal at the end of obstructive apneas was studied (22). In nonrapid eye movement (NREM) sleep, apneas ended with an EEG arousal in 12% and with a movement arousal in the remaining patients, as did all apneas during REM sleep. Associated with obstructive apneas, polygraphic recording shows obvious labored breathing in the ventilatory periods. Paradoxical inward ribcage motion during inspiration can occur throughout REM sleep in children and also occurs during NREM sleep (23). Abdominal muscle recruitment is present during NREM sleep with snoring, and it is inhibited during REM sleep (22). Labored breathing may be the only finding of a polysomnographic recording with or without falls in SaO2 (22,24,25). In such cases, it has been recommended that esophageal pressure swings be monitored during sleep (24,25). Negative esophageal pressures as low as −60 cm H2O can be observed, especially during NREM sleep stages 3 and 4 (25).

**TREATMENT**

Indications for treatment are still under active consideration, taking into account not only obstructive apneas but also respiratory resistive loads during sleep. Treatment should be considered only after establishing the severity of the syndrome with objective testing and after evaluating results from otorhinolaryngologic and maxillofacial studies. Tonsillectomy and adenoidectomy have been the most commonly recommended treatments. However, associated problems such as an abnormally long soft palate, retroposition of the mandible, or soft tissue infiltration behind the base of the tongue must be considered. Such problems may explain residual problems after tonsillectomy. Tracheostomy is currently used only in emergencies. Long-term nasopharyngeal intubation has been proposed and used successfully in infants with Pierre Robin syndrome (26) and other facial dysmorphia (27). Nasal continuous positive airway pressure (nCPAP) should be used more often in children. In younger children, a mask must be custom-made for each child or a commercially avail-
able mask must be modified (28). The indications for maxillofacial surgery are still under discussion. Laryngomalacia can be treated with epiglottoplasty (29). Long-term follow-up examination of infants and children who have been treated for sleep-related upper airway symptoms must be thorough and must include clinical examination, precise questionnaires and polysomnographic recordings, when necessary. OSAS may recur after puberty, especially in males (30). The usefulness of orthodontic treatment of children with mandibular abnormalities after tonsillectomy is still under investigation. Experimental data have shown that disturbed growth of the mandible may occur after prolonged nasal airway blockage during the early period of life (31).

CONCLUSIONS

In summary, sleep-related increases in respiratory resistive loads with or without obstructive apneas can be diagnosed appropriately by pediatricians. A thorough investigation of the anatomic abnormalities involved must be performed to select the most appropriate therapy. Finally, we must be aware of OSAS in members of the same family, including infants, children and parents, with a possible association with sudden infant death syndrome (32).

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