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State of the Art Review

Obstructive Sleep Apnea Syndrome and Its Treatment in Children: Areas of Agreement and Controversy

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INTRODUCTION

Obstructive sleep apnea syndrome (OSAS) has been described in children and teenagers.[1,2] This syndrome has been associated with a series of daytime and nighttime signs and symptoms that may not be apparent at an initial evaluation. The daytime symptoms include excessive daytime sleepiness (EDS), which may be so severe that school authorities become concerned, and abnormal daytime behavior, ranging from signs of aggressivity and h[illegible]activity to pathologic shyness and social withdrawal. Children may exhibit bizarre behavior and have learning problems, morning headaches, frequent upper airway infections, failure to thrive, or obesity. Nocturnal symptoms include breathing difficulty while asleep, heavy snoring, apneic episodes, restless sleep, heavy sweating, nightmares, night terrors, and enuresis.[1–4]

Reasons for seeking consultation tend to vary with age. In children younger than 5 years old, difficult breathing while asleep, heavy snoring, apneic episodes during sleep observed by parents, restless sleep, nightmares, and night terrors are more frequently the reasons for consultation than in older children. This may be so because the parents check young children in their sleep more often than older ones and because young children fall asleep earlier, so that parents have a greater chance to note abnormal sleep patterns. In children older than 5 years of age, EDS (associated with complaints of tiredness and daytime fatigue), abnormal daytime behavior, learning disabilities, frequent morning headaches, nocturnal enuresis, and major discipline problems are more common reasons for consultation.

Failure to thrive and recurrent upper airway infections, both associated with heavy snoring at night, bring children of any age group in for consultation. Reliable data on [illegible] incidence of OSAS are not available.

THE POLYSOMNOGRAM

If OSAS is suspected, an objective evaluation to determine the severity of the syndrome is necessary. A nocturnal polysomnogram, the key diagnostic test, generally includes an electroencephalogram (EEG) (C3/A2–C4/A1 of the 10–20 international electrode placement system), electrocardiogram (ECG), (modified V2 lead), electrooculogram, and chin electromyogram. In young children, adding one or two other EEG channels, such as 03/04 and either F3-F4 or Fp1-Fp2, is helpful in scoring sleep stages. Respiration is monitored using calibrated or noncalibrated respiratory inductive plethysmography in conjunction with airflow monitoring by nasal and buccal thermistors or expired carbon dioxide measurements. Generally, arterial oxygen saturation rather than transcutaneous oxygen tension is monitored. The presence of repetitive obstructive apnea during sleep, associated with oxygen desaturation, is the key objective finding. However, depending on the child's age, the limits of normalcy are not as clearly delineated as one would like. Several issues must be considered when performing this key diagnostic test.

Monitoring has become more accurate with the development of new equipment. Respiratory inductive plethysmography was plagued initially by movement artifacts, and young children tended to disconnect the wires to the

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thoracic and abdominal bands. Based on the original principle of respiratory inductive plethysmography, new systems have been developed that avoid these problems. We have found the Vitalog™ respiratory sensors and "Vitalog™ interface" equipment, which are not overly sensitive to movement, to be helpful in monitoring young infants. Ear oximetry (Biox™) has been useful in older children, but we cannot use this technique in young children, whose ear lobes are very small. In children with dark skin pigmentation, results can be corrected. Finger oximetry has not been as helpful as we had initially hoped. Most finger oximeters had been painful to wear as the night progressed and led to sleep disturbance. However, technical improvement can be expected. Pulse oximetry measured on the finger has given valid results. The response time of transcutaneous electrodes is slower than that of other oximetric equipment. Even with these drawbacks, however, it is possible to monitor oxygen saturation or tension noninvasively.

The nocturnal polygraphic recording should be done with the most accurate equipment available—and that least likely to disrupt sleep. When incomplete obstructions (hypopneas) are suspected, using an invasive technique that may disrupt sleep (esophageal balloon or pressure transducer) is justified on a second night of polygraphic recording.

RESULTS OF POLYSOMNOGRAPHY

The nocturnal polygraphic recording provides information on a number of sleep-related parameters: the number and duration of complete or partial obstructions per hour of sleep, the lowest oxygen saturation during each event, the time spent below a given level of oxygen saturation during the night, the presence and type of cardiac arrhythmias, and the presence and severity of respiratory disturbances, with their impact on the cardiovascular system. It also provides information on the severity of sleep disruption. We have found that scoring sleep in 20- or 30-second epochs, following the international criteria outlined by Rechtschaffen and Kales, may not detect short arousals. Several research teams, including ours, have scored short arousals of up to 10 seconds' duration after an apneic event in order to obtain a better indication of the amount of sleep disturbance. Patients with moderately severe to severe OSAS usually have highly disturbed sleep states, with considerably more stage 1 non-REM sleep, somewhat more stage 2 non-REM sleep, and considerably less (sometimes none) stages 3 or 4 non-REM sleep. REM sleep is often fragmented and of shorter duration than in normal subjects, with a reduction in both total amount of time and percentage of total sleep time. Patients with moderate OSAS have better sleep structure in the presence of stages 3 and 4 non-REM sleep. (Complete apneas are infrequent, usually not occurring during delta sleep.) Regardless of the severity of the syndrome, evaluating miniarousals often gives a better picture of the sleep disturbance than any other sleep score.

When should a respiratory disturbance index be considered pathologic and when should treatment be considered? The Academy of Pediatrics considers an apnea lasting longer than 20 seconds as pathologic in infants, but this definition is not applicable in older children.

Four studies have examined the frequency and characteristics of apneas during sleep in healthy children and teenagers. The total number of children reported was 84, and their ages varied from 2 to 16 years, but few children were younger than 6 or older than 12 years of age. Carskadon et al[5] and Guilhaume et al[6] considered apneas of > 5 and > 10 seconds duration. For apneas longer than 5 seconds in duration, Carskadon et al[5] found a mean of 19 ($\pm$ 8.5) in girls and 17 ($\pm$ 11.5) in boys during total nocturnal sleep time, while Guilhaume et al found a mean of 25 $\pm$ 16. When apneas lasting longer than 10 seconds were measured, Carskadon et al, Guilleminault et al[2] and Tabachnick et al[7] measured a maximum apnea index varying between 3 and 5.5, with all apneas lasting less than 30 seconds, all of them comprising the "central" type, and more than 50% of them seen during stage 1 non-REM sleep.

The long-term pathologic significance of an apnea index between 5 and 10 is not known for either children or adults. Also, the association of tachypnea with rare occurrence or absence of apnea during sleep with or without a decrease of oxygen saturation is infrequently discussed as a marker of pathologic breathing during sleep. While obstructive sleep apnea is always considered a pathologic finding in a polysomnogram, the interpretation of the polysomnographic findings may be difficult when obstructive events during sleep are rare and isolated.

OBSTRUCTIVE APNEA AND OBSTRUCTED AIRWAY DURING SLEEP

Obstructive apneas are defined as persistence (and often a progressive increase) of diaphragmatic movement when airflow ceases at the nose and mouth; they result from an obstruction in the upper airway. OSAS can occur in a normal airway if there are abnormalities in the series of reflexes that control the tension of upper airway dilator muscles; if the drive to the diaphragm and accessory inspiratory muscles either exceeds or precedes that of the upper airway muscles; or if the floppy muscles are "sucked in" during inspiration by negative introthoracic pressure, which meets no opposition from upper airway muscle tension.[8,9] Cherniack and coworkers[10] hypothesized that changes in arterial blood gases can induce these changes in upper airway muscle tension; they pro-

ed a mathematical model of how these blood gas changes can induce apnea.[11] It is possible that insults to certain parts of the brainstem or high cervical cord may have similar results, i.e., abnormal timing between the upper airway muscle and diaphragm contractions.

Nevertheless, OSAS in children is most frequently associated with an upper airway anatomic abnormality. The small airway space, rate of airflow, amount of air intake, intensity and frequency of diaphragmatic and respiratory accessory muscle activity, timing of respiration, body position, state of alertness, and other factors interact over a period of time before a partial obstruction or complete apnea develops. Anatomic abnormalities may induce the initial changes, but reflexes and central nervous system reactions secondary to the anatomic changes must occur for a full-scale OSAS to develop.

The causes of anatomic upper airway abnormalities are numerous, including bone malformations, diseases involving the maxilla and mandible, soft tissue infiltration (which could be related to an endocrine problem, a storage disease, or an infectious disease) and neurologic lesions limiting and impairing muscle contractions (e.g., laryngeal recurrent nerve palsy, bulbo-syringomyelia, palsy involving the IX, X, XII cranial nerves, and poliomyelitis). Finally, poor coordination and timing of the rious reflexes and muscles involved in inspiration and piration can lead to OSAS. As Longobardo and coworkers[11] have shown (see above), this could be secondary to hypoxia, to hypercapnia, or to a respiratory disorder that persists during sleep.

Central apneas, abrupt cessations of diaphragmatic effort resulting in interrupted airflow, are thought to be related to a primary central nervous system derangement. In infants and young children, the possibility that an obstructive mechanism can lead to central apnea is often ignored, thus a CNS defect may be the only cause considered. However, central apnea can also be the result of low lung volume, which can occur in obese children, or children with serious lung disease, when they are asleep in a supine position.

Another cause of central apnea is related to a partial obstruction of the upper airway. We studied five infants with Pierre Robin syndrome who initially had central apneas that, as the children matured, disappeared and were replaced by mixed and obstructive events.[12] We have also followed five children between 7 and 14 years of age who had clear retrognathia and an abnormally small hypopharyngeal airway passage (3 to 5 mm in diameter) during wakefulness, documented by cephalometric roentgenograms. Polygraphic investigation of these children identified a mean total of 168 central apas (range 92–229) during sleep. Because there appeared to be a discrepancy between the finding of central apnea and the presence of intermittent loud snoring, each child was monitored again during sleep with an esophageal balloon or a pressure transducer. This indicated an increase of esophageal pressure (to 40 cm H_2O), particularly during REM sleep, just before the appearance of central apnea. The mechanism(s) responsible for the abrupt development of central apnea following partial occlusion of the upper airway during sleep is unclear.[13] One possibility has been recently emphasized by Dempsey and Skatrud in adults.[14] These authors have shown that snorers who often have an upper airway narrowing during sleep have an airway resistance that is ten times higher at sleep onset than during wakefulness. This is compared with a two- to four-fold increase in normal subjects. They have also shown that hypoxia during sleep may lead to respiratory instability, with the development of post-hyperventilation central apnea when a cluster of hyperpneic breaths is so large that it decreases P_{CO_2} below a sleep-related "apneic threshold" (which is at a higher level than during wakefulness). This is only one possible explanation, but the fact remains that a partially obstructed airway may be associated with central apnea. The use of nasal continuous positive airway pressure (CPAP) as a transient testing procedure, and in one case, maxillofacial surgery,[15] indicated that affecting the upper airway narrowing eliminated this type of central sleep apnea. These observations demonstrate that a partial obstruction of the upper airway, such as by mandibular malformation in our cases, can be associated with central apnea during sleep.

SNORING WITHOUT OBSTRUCTIVE APNEA

A partially obstructed airway during sleep may lead to clinical symptoms, frequently associated with obstructive sleep apnea syndrome, but without evidence of apnea. We have reported on several children who experienced sleepiness during the day and loud snoring, heavy sweating, and restless sleep at night, but who had no obstructive apneas,[16] or only very few central, mixed, and/or obstructive events in combination during REM sleep. Objective daytime testing with the multiple sleep latency test, documented that the children were sleepy.[17] Monitoring with an esophageal balloon or pressure transducer indicated that esophageal pressure was significantly higher (20–40 cm H_2O) than expected in comparison with normal control values, particularly during REM sleep. There was an upper airway load, most commonly due to enlarged tonsils and adenoids, which caused clinical symptoms similar to those of OSAS. However, no oxygen desaturation was noted during the nocturnal polygraphic recording, and no repetitive EEG arousals were monitored. The laborious breathing was behaviorally observable, and a significant tachypnea during sleep was the major polygraphic finding during sleep.

These cases indicate that, in children, a history of loud, repetitive heavy snoring that has been present every night for several months should be clinically investigated.

CLINICAL EXAMINATION

Regardless of the combination of symptoms, every patient should be evaluated thoroughly. A careful history should be taken to determine if the OSAS is secondary to another syndrome that is responsible, at least partially, for the sleep-related upper airway occlusion.[18]

The following should be investigated in an evaluation of the oro-naso-maxillo-facial region: the aspect of the nares, the collapse of the nostrils with inspiration, the size of the nose, the tongue's size and consistency, the length of the tongue's protrusion, the width of the mouth, the length and position of the soft palate, the presence of an abnormal amount of lymophoid tissue, the enlargement of tonsils and adenoids, the presence of retrognathia, micrognathia, cleft palate or scars from its repair, infiltration of oropharyngeal soft tissues by storage disease, and so on. If there is any doubt, heavy chronic loud snorers, as well as children with repetitive obstructive sleep apnea, will benefit from upper airway investigation while awake.

IMAGING TECHNIQUES AND THE UPPER AIRWAY

It is important to obtain a clear picture of the configuration of the upper airway, which can be accomplished with imaging techniques. All techniques used to investigate OSAS in adults can also be used in adolescents. Three are particularly helpful: cephalometric roentgenograms,[19] computerized tomography scans,[20] and fluoroscopy during sleep.[21,22] A fourth technique, magnetic resonance imaging, is also useful but expensive. In deciding which technology to use, one must weigh the amount of information provided by the expense of the procedure. Each of the four techniques can be useful, and all have some drawbacks; therefore the device must be made on an individual basis.

Cephalometric roentgenograms are inexpensive and quickly obtained, but the radiologic laboratory must have the equipment (i.e., Wermer Cephalo-stat) required to stabilize the child's head in the appropriate position and must produce roentgenograms that outline soft tissues so that facial contours appear on the film. The major impediments to its general use are that many radiology laboratories do not produce sufficiently clear films and that interpreting the infiltration of soft tissue into the airway requires careful training.

Computerized tomographic scans are more expensive. Often a longitudinal "scout" cut will provide most of the information needed. The horizontal cuts appraise the relationship of the various anatomic structures to the airway space.

Fluoroscopy during sleep provides for a dynamic investigation of the upper airway, but requires that the child sleep on a table in the laboratory. The patient should also be monitored polygraphically during fluoroscopy, so that sleep states can be identified.

Although it is the most expensive method, we have also used magnetic resonance imaging (MRI), to avoid large exposure to radiation, with "gating" of signals as a function of respiratory movements (i.e., inspiration and expiration).

In very young children, computerized tomographic scans and MRI may be easier to perform during sleep, especially non-REM sleep. For both techniques the head must be immobilized, with the nose at a 90° angle in relation to the plane of the radiologic table; ear bars, or plastic head fixaters for MRI, similar to those for cephalometric roentgenograms, help in steadying the head, thus facilitating a comparison of scans of the same region. It is important to correlate these images with phases of respiration and swallowing, which radically alter the configuration of the orohypopharyngeal space, which is a dynamically active region.

Choosing the correct technology is most difficult in young children, particularly if OSAS is associated with mental retardation, for example in cases of Down's syndrome or Crouzon's disease. We have little normative data for cephalometric roentgenograms for children between 12 months and 5 years of age, and no studies of systematic computerized tomographic scans or fluoroscopic evaluation in this age group have been reported.

Imaging techniques should always be used in association with pharyngolaryngological evaluation, and also with a fiberoptic endoscopy performed on a supine child. If possible, depending on the child's cooperation, specific movements of the jaw (e.g., prognathic displacement) or maneuvers (Valsalva or Muller) should be performed during the fiberoptic evaluation.

EVALUATION OF RISKS ASSOCIATED WITH OBSTRUCTIVE SLEEP APNEA

Hemodynamic Changes

There are no reports of nocturnal hemodynamic studies in children similar to those conducted in adults, with arterial lines to continuously monitor nocturnal pulmonary arterial pressure, systemic arterial pressure, cardiac output, etc. In early descriptions of OSAS in children the presence of high blood pressure was noted. Numerous tests for other causes of the increased systemic pressure were negative, and after tracheostomy the systemic blood pressure values returned to normal, both early and at the 10-year posttracheostomy evaluation. The most commonly noted cardiovascular problem in children with

O[illegible] was acute cardiac failure, occasionally the first indication of the syndrome observed by medical personnel. Before OSAS was recognized, several authors had noted a relationship between heavy snoring, enlarged tonsils and adenoids, and cardiac failure.

Several children, mostly prepubertal, were referred to us after hospitalization in intensive care units for acute cardiac failure. For at least a year prior to the cardiac failure, their OSAS was usually unrecognized, despite the presence of daytime and nighttime symptoms. The failure itself often occurred when the child contracted a cold or bronchopneumopathy, which may not have been severe but in combination with the chronic nocturnal problem led to the acute cardiac failure. The OSAS was documented polygraphically after each child's condition stabilized.

Cardiac Arrhythmias

No systematic study of cardiac arrhythmias associated with OSAS in prepubertal children or teenagers has been reported. In our own population, the most common arrhythmia, with the exception of the classic brachytachycardia seen with apnea, were significant sinus brachycardia and sinus arrests. The terminology "extreme sinus brachycardia" refers to heart rate below 35 beats/minute i[illegible]e 2–4-year-old group and below 30 beats/minute in o[illegible]r children. Eight children (8% of our population) had at least one episode of extreme brachycardia while monitored. Another eight (8% of our population) had a sinus arrest lasting between 2.5 and 6 seconds, associated with obstructive apnea, during one night of polygraphic monitoring; five (2.5% of our population) had isolated episodes of second-degree atrioventricular block, also directly associated with an obstructive apnea. No other significant cardiac arrhythmia was seen.

Anoxic Seizures

Nocturnal seizures in combination with OSAS have been more common in our population of children than in adults with OSAS. The majority of our children already had an abnormality of the brain (i.e., cerebral palsy, or a syndrome with a seizure disorder, such as Prader-Willi Syndrome), but nocturnal seizures had never occurred or were controlled. A major reason for seeking medical consultation was that the children's nocturnal seizures had increased in frequency. To treat the increased seizure activity occurring just before bedtime, a 9-year-old boy with Prader-Willi syndrome was administered a larger dosage of barbiturates before going to bed; yet the frequency and severity of the seizures increased. At the polygraphic evaluation of this child, it was apparent that [illegible]s CNS depressant, administered to control epilepsy, had worsened the OSAS and increased nocturnal hypoxemia. Treating the OSAS in all these patients resolved the unexplained worsening of nocturnal seizures.

The evaluation of all these potential risks must be performed before treatment can be considered.

TREATMENT OF OSAS IN CHILDREN

The indications for treatment are not well codified, and certain types of treatment are considered highly controversial in children. One reason for this is the lack of objective data on the long-term effects of some proposed therapeutic procedures. Another one is, as already pointed out, our lack of information on the long-term detrimental effects of mild-to-moderate polygraphic abnormalities. The Stanford Sleep Clinic philosophy, based on our 196 OSAS patients, 2 to 18 years of age, has been:

1. Treatment should be considered only when the severity of the syndrome has been established by objective testing and after results from otorhinolaryngologic and maxillofacial evaluations, including fiberoptic and imaging data, have been reviewed.
2. Once the underlying cause of the OSAS has been established, the most appropriate approach, based on the current state of knowledge, should be tried. The fact that there is an improved understanding of the underlying causes of OSAS, and that new imaging techniques and treatment procedures have been introduced, explains some changes in therapeutic recommendations.

Tonsillectomy and Adenoidectomy

Classically, treatment for OSAS in children and teenagers has been surgical. Tonsillectomy, or tonsillectomy and adenoidectomy (T & A) have been the most commonly recommended.[2–4] Too often, however, not enough attention is paid to associated problems such as an abnormally long soft palate, retroposition of the mandible, or soft tissue infiltration behind the base of the tongue, that may be present with enlarged tonsils and adenoids, and may explain residual apnea after tonsillesctomy. Furthermore, if T & A is performed during the prepubertal years, there is a chance that in puberty, when extensive soft tissue growth occurs in males associated with increased testosterone secretion, the OSAS will reappear particularly in those whose airway space is already compromised by a malocclusion with mild to moderate retroposition of the lower mandible. This has occurred in four of our patients who were seen between six and nine years of age with clinical symptoms of heavy snoring at night with daytime sleepiness. They had obstructive sleep apnea syndrome confirmed by polysomnography, with an apnea index monitored between 31 and 56; a follow-up nocturnal recording, within five months post T & A had demonstrated an apnea index between 4 and 7, but we were recontacted by parents when the children were between 12 and 15 years of age. Heavy snoring at night, without clear clinical complaints, had reoccurred, and

objective nocturnal polygraphic monitoring demonstrated an apnea index between 15 and 28, confirming parental reports.

Tracheostomy

In the past, tracheostomy was a frequent treatment when tonsillectomy and adenoidectomy were insufficient.[23] Tracheostomy always resolves the OSAS, but it may cause secondary problems, such as depression in children following the surgery, and difficulties within the family accepting the surgical solution and caring properly for the stoma. Nevertheless, tracheostomy has been clearly beneficial in many cases.

In the more recent past, following knowledge gained in dealing with adult OSAS, three other treatment modalities have been used in our own patient population: uvulopalato-pharyngo-plasty, maxillofacial surgery, and nasal CPAP.

Uvulopalatopharyngoplasty (UPPP)

This procedure, which Ikematsu[24] originally performed to control snoring, was introduced by Fujita et al[25] as a means of treating adult OSAS. It consists of the removal of the thin posterior half of the soft palate, with excision of some underlying muscle from the tonsillar fossa and of excess lateral posterior wall tissue. Follow-up studies of adult OSAS patients demonstrated that not all subjects were good candidates for this procedure.[26] Samelson,[27] criticizing this surgical treatment, raised the issue of the sequelae and complications of UPPP: in addition to the immediate chance of bleeding, there are more persistent problems—nasal reflux, retraction, and the ensuing reduction of the airway, as well as the possible impact on the vocal trill. Undoubtedly, these complications (with the exception of the undocumented impact on the vocal trill) have occurred in adult patients undergoing UPPP. Therefore, before performing surgery on the upper airway in children, the potential gain versus the potential complications of the procedure must be carefully weighed. We have recommended this procedure in six cases: in five boys and one girl, mean age 4.2 years $\pm$ 10.8 months. These children had developed symptoms of apnea during sleep between 3 weeks and 2½ years of age. Three boys diagnosed with apnea between 3 weeks and 4 months of age were full-term and were considered to have "apnea of infancy" ("near miss" sudden infant death syndrome). The three other children were brought to medical attention for a "breathing problem" during sleep between 14 months and 1½ years of age. All children not only had nocturnal polygraph recordings, but also fiberoptic scope evaluation. Four patients had previous tonsillectomy and adenoidectomy (T & A), and two had undergone an isolated tonsillectomy prior to consideration of UPPP. One patient, after documentation of the poor response to T & A, had a tracheostomy performed, which was closed 2½ months later at the family's request. These children were followed 2–6 years post-UPPP (i.e., some were investigated before systematic evaluation of discrete craniofacial abnormalities by cephalometric X-rays). Comparison on the initial pre-and 4 months post-UPPP polygraphic recordings indicated that, in all cases, the clinical symptoms disappeared and objective improvements occurred in the breathing problem during sleep. The mean apnea index after T & A or tonsillectomy alone was 27.2, and it was 0.05 post-UPPP. In four patients, long-term follow-up indicated a persistent beneficial effect of UPPP, and an absence of any complications. In the last two subjects, no complications were noted, but the parents reported persistent snoring and restless sleep, with upper airway infections, hay fever, and respiratory allergy. Objective polygraphic monitoring performed at the time of an upper airway infection in these two children showed some obstructive events during REM sleep, asociated with mild drops in oxygen saturation (91–90%). These two subjects had cephalometric roentgenograms during the post-UPPP follow-up, which documented malocclusion class II. and smaller-than-expected posterior airway space widths of 5 mm and 6 mm, respectively. They also had a discrete retroposition of a mandible, associated with decreased size of the upper airway behind the base of the tongue. This anatomical abnormality could explain the persistence of mild obstructive symptoms during sleep in association with upper respiratory infections.

Maxillofacial Surgery

Maxillofacial surgery has been used since the 1970s to treat OSAS, but its efficacy has been controversial, because of the risk of relapse. Piecuch[28] reported one child, Kuo and colleagues[29] two cases, and Bear and Priest[30] three cases of OSAS that were resolved by maxillofacial surgery; postsurgical sleep monitoring was not conducted in any of these cases, however.

At Stanford, 130 adult patients with OSAS underwent maxillofacial surgery, were by a different surgical approach from those previously described. More specifically, it combined a hyoid myotomy and resuspension with different types of maxillomandibular osteotomy.[31,32] Of five teenagers included in that group, two had bilateral mandibular osteotomy and inferior sliding osteotomy with hyoid myotomy and resuspension; three others had maxillomandibular osteotomy with inferior sliding osteotomy, hyoid myotomy, and resuspension. These five cases have been followed for a maximum of 3½ years (mean of 25 months). Postsurgical polygraphic monitoring documented that the respiratory disturbance index. the level of oxygen saturation during sleep, and nocturnal sleep disturbances all improved significantly. The respiratory disturbance index was below 10, and nocturnal oxygen saturation was above 94%. No complications were noted.

Nasal Continuous Positive Airway Pressure (CPAP)

Recently we have used nasal CPAP[33] in pubertal and postpubertal patients who required orthodontic preparation before undergoing maxillofacial surgery. As with tracheostomy, nasal CPAP effectively resolves OSAS, regardless of the mechanisms responsible for the problem. We now also use nasal CPAP instead of tracheostomy to treat prepubertal children who are either too young for surgery, or for whom surgery would be inappropriate, or in whom surgery would be too difficult to perform. In trial studies, we used commercially available CPAP equipment (Respironics™) in a group of eight teenagers with no difficulties during a 5-month follow-up period. We had more problems in a group of five prepubertal children.[34] Because the younger children's faces were small or abnormally shaped, we had to build our own masks or modify the commercially available ones; long-term treatment was, however, successful in four out of five patients. The complications and problems associated with nasal CPAP have been related to: 1) the children's difficulties in understanding how the mask and CPAP equipment function (many were mentally retarded); 2) the parents' difficulties collaborating with the medical team in training the child to keep the nasal mask his/her face; 3) air leaks at the edge of the mask, causing eye irritation and leading to the reappearance of apnea; and 4) skin allergy to the cement used to initially fix the masks in small children. Problems 1 and 2 were responsible for one child's abandoning nasal CPAP treatment; problems 3 and 4 were occasionally bothersome, but never led to interruption of therapy. The theoretical risks of stomach dilatation due to incorrect administration of CPAP never occurred either in children or in adults under chronic nasal CPAP. (Over 150 were followed for over 15 months.) The development of unspecified rhinitis, secondary to nasal CPAP, has been a complication in 20–25% of our adult patients. Our population of children using nasal CPAP is still small, and more research is needed to refine this therapy in younger age groups.

Induced Weight Loss

Obesity is a relatively rare problem in pubertal and adolescent patients with OSAS, as compared with its incidence in adults; it is sometimes seen in association with metabolic problems related to another syndrome. Failure to thrive is seen more often. If obesity is a problem, then induced weight loss may be helpful.

Pharmacologic Treatment

Pharmacologic treatment, particularly with tricyclic medications (protriptyline) and medroxyprogesterone, are rarely used in children. Orenstein and coworkers[35] reported using progesterone in one patient. This drug acts on obesity-related hypoventilation during sleep as part of the restrictive chest bellow disease, rather than on the upper airway obstruction. Medroxyprogesterone affects glomerular filtration rate and serves as an antialdosteronic diuretic. It may be helpful in obese children who have a combination of problems, but it is not a major component in the treatment of OSAS, per se. Protriptyline and imipramine may be helpful, but there are no long-term reports on efficacy and complications in children with OSAS.

CONCLUSION

This review indicated that many therapeutic avenues may be available to treat children and teenagers with OSAS. It is, therefore, critical to carefully evaluate the different components leading to OSAS. Obviously, anatomic abnormalities play a role and must be identified. However, other factors, such as the child's age, teeth, facial structure, and intellectual development, and the presence of mental retardation or the possible association with other congenital anomalies or syndromes, etc. must also be investigated before deciding upon a given treatment.

At Stanford, we have tried to eliminate tracheostomy as a therapeutic approach and have avoided pharmacologic treatments that have not been overly successful in adults. These are our biases. Undoubtedly, tonsillectomy, or T & A are still the first choices in the treatment of "uncomplicated" OSAS in early childhood. However, long soft palates, small mandibles, and low-positioned hyoid bones immediately attract our attention, and efforts are made to follow affected children for long periods of time, or at least through puberty, by objective testing during sleep, in order to evaluate the eventual need for further surgery. Failure of T & A does not automatically lead to further surgery. Nasal CPAP had been well tolerated by a 3-year-old mentally retarded child and a 5-year-old Down syndrome child. The advantage of nasal CPAP is that it immediately controls the OSAS, providing time for reevaluating the child during physiological and emotional growth, and for planning surgery, also taking into consideration teeth, bone growth, etc. Obviously, it is irrational to consider nasal CPAP as the only treatment of OSAS in a young child when a surgical treatment could eliminate the problem, but it may be, at times, the best solution for heavily retarded children. Finally, there is an issue not currently being explored: recent systematic studies in adults have shown that moderate craniofacial anomalies are excessively common in adult OSAS and contribute to the development of the syndrome. The question arises: could orthodontic treatment of prepubertal children with mandibular abnormality allow growth of the mandible and avoid further worsening of OSAS during the pubertal period when the tongue muscles enlarge? This attractive thought is under

investigation at Stanford, but currently it is not yet supported by long-term follow-up data.

In conclusion, OSAS can be diagnosed appropriately with polygraphic techniques by pediatricians. However, there is considerable uncertainty concerning the long-term significance of mild-to-moderate polygraphic respiratory abnormalities in the absence of clear clinical complaints. On the opposite side of the spectrum, a significant increase in airway resistance during sleep without apnea may lead to as many clinical complaints as a typical OSAS and should be treated adequately. There are many mechanisms that can lead to upper airway obstructions during sleep; pediatricians should become aware of them. New treatments for OSAS are available, so that a permanent tracheostomy is no longer the only option for patients with severe OSAS. However, a thorough investigation of the anatomic abnormalities involved in the development of OSAS must be performed to select the most appropriate therapy. Finally, all children treated for OSAS should have regular follow-up examinations, with testing during sleep.

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