Use of nasal continuous positive airway pressure as treatment of childhood obstructive sleep apnea

Carole L. Marcus, MBBCh, Sally L. Davidson Ward, MD, George B. Mallory, MD, Carol L. Rosen, MD, Robert C. Beckerman, MD, Debra E. Weese-Mayer, MD, Robert T. Brouillette, MD, Ha T. Trang, MD, PhD, and Lee J. Brooks, MD

From Johns Hopkins University, Baltimore, Maryland; Childrens Hospital Los Angeles, Los Angeles, California; St. Louis Children's Hospital, St. Louis, Missouri; Yale University School of Medicine, New Haven, Connecticut; Tulane University Medical Center, New Orleans, Louisiana; Rush Presbyterian-St. Luke's Medical Center, Rush University, Chicago, Illinois; McGill University, Montreal, Quebec, Canada; Hôpital Antoine Beclere, Clamart, France; and Rainbow Babies and Childrens Hospital, Cleveland, Ohio

Objective: **To determine the safety and efficacy of nasal continuous positive airway pressure (CPAP) for obstructive sleep apnea (OSA) during childhood and the effects of growth and maturation on CPAP requirements.**
Design: **Retrospective study with use of a written questionnaire administered to pediatric practitioners treating sleep disorders.**
Setting: **Nine academic pediatric sleep disorders centers.**
Results: **Data were obtained for 94 patients. Three percent of patients receiving CPAP were less than 1 year, 29% were 1 to 5 years, 36% were 6 to 12 years, and 32% were 13 to 19 years of age; 64% were boys. The longest duration of CPAP use was 4 years. Indications for CPAP included OSA associated with obesity (27%), craniofacial anomalies (25%), idiopathic OSA persisting after adenoidectomy and tonsillectomy (17%), and trisomy 21 (13%). Continuous positive airway pressure was effective in 81 patients (86%), in one patient it was unsuccessful, and in 12 patients compliance was inadequate. The median pressure required was 8 cm H_2O (range, 4 to 20 cm H_2O); pressure requirements were independent of age or diagnosis. Twenty-two percent of patients eventually required a modification of CPAP levels. Complications of CPAP were minor. Sixty-four percent of centers reported difficulty in obtaining funding for CPAP.**
Conclusions: **Continuous positive airway pressure is safe, effective, and well tolerated by children and adolescents with OSA. Experience in infants is limited. As pressure requirements change with patient growth, we recommend that CPAP requirements be regularly reevaluated over time. The marked center-to-center variability in CPAP use suggests that specific indications for this therapy require clarification. (J PEDIATR 1995;127:88-94)**

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Reprint requests: Carole L. Marcus, MBBCh, Johns Hopkins Hospital, Eudowood Division of Pediatric Respiratory Sciences, Park 316, 600 N. Wolfe St., Baltimore, MD 21287-2533.

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Obstructive sleep apnea is a significant cause of morbidity in children. Complications include failure to thrive, cor pulmonale, developmental delay, and even sudden death.[1-3] The majority of children with OSA have adenotonsillar hypertrophy and can be treated successfully by tonsillectomy and adenoidectomy.[3] However, a small percentage of these children have persistent OSA after surgery and require further intervention. Furthermore, children with other predisposing factors for OSA (e.g., craniofacial anomalies, neuromuscular weakness, or obesity) may require alternative treatment. Until recently, tracheostomy was the primary treatment for this group of patients and is still the only treatment offered at many institutions.

During the past 15 years, nasal continuous positive airway pressure has been used widely in the treatment of adults with OSA.[4] However, other than one early report by Guilleminault et al.,[5] reports on the use of CPAP to treat OSA in children have been limited to case reports or abstracts.[6-10] The current commercial availability of CPAP, including the availability of pediatric-sized masks, has resulted in more widespread use in children; thus the report by Guilleminault et al. may not be representative of current practices. Some fundamental questions regarding the use of CPAP in children compared with the use in adults have yet to be answered. Specific areas of interest include the efficacy of CPAP during childhood, the level of airway pressure required, the frequency of complications, the effect of growth on CPAP requirements, and the degree of compliance that may be expected.

CPAP	Continuous positive airway pressure
OSA	Obstructive sleep apnea

Because the numbers of children with OSA undergoing CPAP therapy are small, we pooled the data from pediatric centers with expertise in sleep-disordered breathing and present a retrospective review of the use of CPAP in the treatment of childhood OSA.

METHODS

Retrospective data on the use of CPAP in children were obtained from detailed questionnaires sent to 11 major pediatric sleep disorder centers (nine in the United States, one in Canada, and one in France). The questionnaires were answered by the medical directors. When necessary, further information was obtained by telephone. Patients using CPAP or bilevel positive airway pressure (BiPAP, Respironics Inc., Murrysville, Pa.) (which provides different levels of inspiratory and expiratory pressure) for conditions other than OSA (e.g., respiratory failure) were not included. Variables between groups were compared by analysis of variance.

Table. Predisposing conditions for OSA in 94 children

Predisposing condition	n (%)
Obesity	25 (27)
Craniofacial anomalies*	23 (25)
Idiopathic (total)	17 (18)
T & A performed	16 (17)
T & A not performed†	2 (2)
Trisomy 21	12 (13)
Neuromuscular disease	5 (5)
Mental retardation and cerebral palsy	5 (5)
Pharyngeal flap surgery	4 (4)
Arnold-Chiari malformation	2 (2)
Other‡	1 (1)
Pending T & A	1 (1)

T & A, Tonsillectomy and adenoidectomy
*Chromosomal anomalies (n = 3), Pierre Robin syndrome (n = 3), achondroplasia (n = 2), Treacher Collins syndrome (n = 2), pyknodysostosis (n = 2), maxillary hypoplasia (n = 2), Crouzon syndrome, Binder syndrome, incontinentia pigmentosa with maxillary hypoplasia, Apert syndrome, congenitally small nasopharynx, fibrous dysplasia of the mandible.
†Includes patient with cystic fibrosis.
‡Tracheomalacia after tracheal reconstruction in child who had tracheostomy because of laryngeal papillomatosis.

RESULTS

Responses were received from nine of the 11 centers. The remaining two centers were unable to furnish detail ed information because of lack of records and were therefore not included in the study. Of these two centers, one reported following only a few patients; the other stated that it followed a large group of patients but could not provide exact numbers.

Patient demographics. Data were obtained for 94 patients. Three percent of the patients were less than 1 year, 29% were 1 to 5 years, 36% were 6 to 12 years, and 32% were 13 to 19 years of age; 64% were boys. The median duration of follow-up was 13 months (range, 1 to 48 months). Indications for the use of CPAP are shown in the Table.

Institution of CPAP therapy. All centers reported using similar indications for the institution of CPAP therapy. Diagnosis of OSA was by polysomnography. All centers referred patients with enlarged tonsils or adenoids for surgery before CPAP therapy was considered; in the majority of these patients abnormalities resolved after tonsillectomy and adenoidectomy. Use of CPAP was reserved for those patients who had symptoms (e.g., daytime hypersomnolence, poor growth, or pulmonary hypertension) and had polysomnographic abnormalities (obstructive apnea, arterial oxygen desaturation, and hypercapnia), in whom tonsillectomy and adenoidectomy were ineffective or not indicated. In the majority of cases (76%) CPAP was instituted as a second-line treatment after tonsillectomy and adenoidectomy. Only one center frequently used CPAP as a first-line

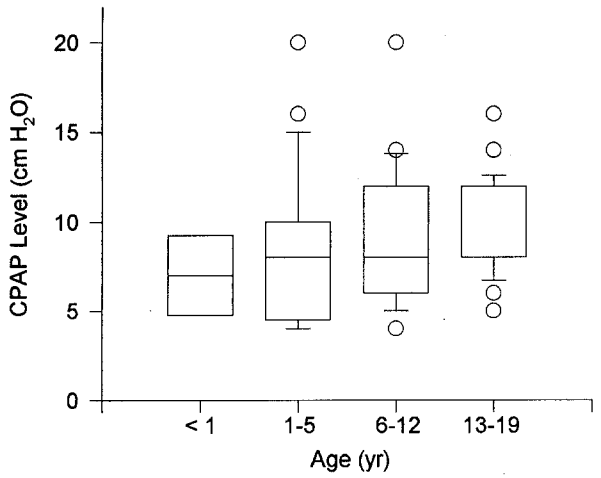

Fig. 1. Levels of *CPAP* according to age group, are shown for 70 subjects for whom levels were available. *Lower boundary* of box indicates 25th percentile of range, *upper boundary* indicates 75th percentile, and *horizontal line* within box indicates median. *Error bars* depict 10th to 90th percentiles; data points outside this range are shown as *circles*. No differences were found in pressure requirements between different age groups.

treatment in children with OSA who did not have enlarged tonsils or adenoids (by physical examination and lateral neck x-ray studies or endoscopic evaluation). The other centers advocated tonsillectomy and adenoidectomy as initial treatment for most patients even if the tonsils and adenoids were not very large, or if the child had other predisposing conditions for OSA, with the rationale that any degree of airway encroachment by the tonsils and adenoids would be detrimental. Exceptions to this were patients who were at increased surgical risk (e.g., morbidly obese patients) or those who had minimal tonsillar or adenoidal tissue. Most centers performed initial diagnostic polysomnograms, followed by repeated overnight polysomnography during which CPAP levels were titrated. However, one center instituted CPAP therapy during the initial polysomnogram, another center instituted CPAP during nap polysomnography, and a third instituted CPAP therapy on the inpatient ward and then proceeded with polysomnography. One center titrated pressure levels with bilevel positive airway pressure (BiPAP) in the laboratory but then discharged patients home on a CPAP regimen. Six of the nine centers instituted CPAP therapy as an inpatient procedure. In most centers various medical personnel, including physicians, nurses, respiratory therapists, polysomnography technicians, and home care company personnel, were responsible for training the child and family in the use of CPAP. One center, which relied primarily on outpatient CPAP training, used behavioral psychologists to help train young or developmentally delayed patients.

Equipment. Several different CPAP machines were used. The manufacturers of the machines used most frequently included Respironics Inc. (Murrysville, Pa.), Healthdyne Inc. (Marietta, Ga.), Puritan-Bennett Corp. (Overland Park, Kan.), and Medtronic Inc. (Minneapolis, Minn.). No center thought that a particular CPAP model was superior. Similarly, centers reported successful experience with a variety of nasal masks; the ones used most often were the Respironics and Sullivan (Rescare Ltd., Sydney, Australia) masks. Although many children with craniofacial anomalies were using CPAP, only two patients required custom-made masks. Five of nine centers reported occasional or frequent difficulty in obtaining funding for CPAP equipment. Funding difficulties were encountered when patients were insured by private insurance companies, health maintenance organizations, and governmental medical assistance. Four centers reported the use of bilevel positive airway pressure (BiPAP) rather than CPAP in selected patients with OSA, because it was usually better tolerated than CPAP, but reported frequent difficulty in obtaining funding for the equipment. One center used the BiPAP machine in the laboratory situation but had not found it useful in the home. The remaining four centers had no experience with the BiPAP machine. Patients underwent cardiorespiratory monitoring rarely, because these monitors do not detect obstructive apnea.

Pressure requirements. Seven centers were able to provide CPAP levels for each patient; these data were not documented at the other two centers. Therefore the exact CPAP levels were available for 70 subjects. No differences in patient demographics were observed between the seven centers that provided data and the remaining two centers. The median CPAP level required to overcome obstructive apnea, arterial oxygen desaturation, and hypercapnia was 8 cm H_2O (range, 4 to 20 cm H_2O). Ten patients required supplemental oxygen in addition to CPAP to maintain oxygenation at tolerable CPAP levels. No significant differences in CPAP levels were observed among the different age groups (Fig. 1); the correlation between CPAP level and age was 0.09 (not significant). No differences were observed in pressure levels among children with obesity, craniofacial anomalies, or idiopathic OSA. Because of the different techniques used in the performance, scoring, and reporting of polysomnography, we were unable to compare pressure levels with the severity of OSA. Most centers reexamined patients with polysomnography (or in two cases, home oxygen saturation recordings) every 4 to 12 months. Two centers reexamined patients with polysomnography only if they had symptoms. Treatment with CPAP was considered to be effective if it resulted in resolution of clinical symptoms, normoxia during sleep, and a marked improvement in other polysom-

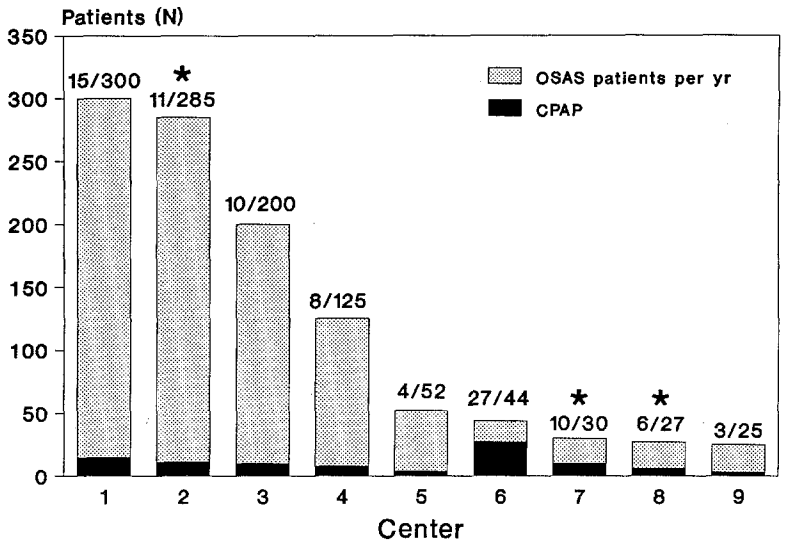

Fig. 2. Number of patients at each center who are currently using CPAP and number of patients in whom OSA is diagnosed annually. * Center does not evaluate uncomplicated cases of suspected OSA caused by adenotonsillar hypertrophy.

nographic abnormalities. Of the patients with adequate compliance, CPAP was effective in treating OSA in all but one. Three patients with tracheostomies successfully underwent decannulation after the institution of CPAP therapy. Twenty-one patients (22%) required changes in their CPAP pressure with time. Use of CPAP was discontinued in four patients in whom OSA resolved, and pressure levels were decreased in another four patients. In three of the patients in whom pressure levels were decreased, the levels were decreased after airway surgery (craniofacial surgery, or tonsillectomy and adenoidectomy); only one patient had a spontaneous decrease in pressure needs. No patient had decreased CPAP requirements as a result of weight loss. However, only two patients were able to lose weight, and this weight loss was transient. Thirteen patients (14%) required an increase in CPAP with time. In five of these patients the increase was associated with weight gain and in another with airway granulomas.

Side effects. Frequent side effects included nasal symptoms (congestion, dryness, or rhinorrhea) and symptoms related to poor mask fit (eye irritation, conjunctivitis, dermatitis, or skin ulceration). No episodes of pneumothorax, abdominal distention, emesis, aspiration, or clinical evidence of decreased cardiac output were reported. Theoretically, children may be at increased risk for otologic complications because of their predisposition to otitis media. However, ruptured tympanic membranes or tinnitus was not reported, although four patients at one center were noted to have erythematous tympanic membranes. Use of CPAP was reported to increase oral secretions in children with mental retardation. One patient reported headache. All symptoms responded to therapy, and in no case did symptoms necessitate discontinuation of CPAP.

Compliance. Compliance was described in terms of CPAP use as a percentage of hours prescribed, with compliance considered to be adequate if CPAP was used for at least 50% of the hours prescribed.[11] Most centers obtained readings of the equipment hour meter to verify parental reports of compliance. However, some equipment did not have hour meters; in other cases personnel were unavailable to examine the equipment. Most centers reported an estimated compliance rate of 50% to 100%, and several centers reported an estimated compliance rate of 75% to 100%. Adolescents were reported to be the least compliant. Measures used to enhance compliance included modification of the equipment (e.g., using a "ramp" function that gradually increased the airway pressure during a set time, finding a mask with an improved fit, adding a humidifier, or adding a low-pressure alarm), frequent clinic visits, and professional counseling. In one case of parental neglect, child welfare officials were involved in achieving compliance. No patient required arm restraints. Poor compliance was thought to result in inadequate treatment in a total of 12 patients (13%). In 50% of these cases compliance problems were attributed primarily to the parent rather than to the child.

Variability in CPAP use among different centers. The number of patients using CPAP at each center varied widely, ranging from three to 27 patients (median, 10 patients) (Fig. 2). The number of patients in whom OSA was diagnosed annually at each center varied from 25 to 300, and the pro-

portion of patients using CPAP, as a percentage of those in whom OSA was diagnosed annually, varied from 4% to 61%. Centers used similar criteria for instituting CPAP; all reported that the decision to implement CPAP therapy was based on clinical symptoms together with polysomnographic criteria in children with OSA. All centers recommended tonsillectomy and adenoidectomy as the first line of treatment in children with OSA and adenotonsillar hypertrophy. Morbidity and mortality resulting from OSA were similar among the centers. Only one death occurred in a patient with OSA; this was related to anesthetic complications (stiff chest syndrome after fentanyl administration[12]). One center (No. 9), which had few patients using CPAP, had referred five patients with OSA for tracheostomy within the past year; the remaining eight centers had referred up to two patients each for tracheostomy. Centers reported up to two patients each with cor pulmonale or failure to thrive persisting after treatment of OSA. No patient receiving CPAP had persistent cor pulmonale or failure to thrive.

Part of the difference in CPAP use among the centers could be attributed to differences in the patient populations. Although most centers examined all patients with suspected OSA, three centers did not routinely examine patients with uncomplicated OSA caused by adenotonsillar hypertrophy. One center (No. 2) used CPAP therapy frequently in children with OSA caused by craniofacial anomalies (9/11 patients receiving CPAP), whereas center 6 was the only center using CPAP in children with OSA related to mental retardation and cerebral palsy (5/27 patients receiving CPAP). Center 8 stated that difficulty in obtaining funding for polysomnography and CPAP equipment was a major obstacle to CPAP use. In addition, the date of introduction of CPAP at the various institutions varied from 1985 to 1993; center 6, which began using CPAP earlier than the other institutions, had the largest accumulation of CPAP patients.

Because CPAP is the mainstay of treatment for adults with OSA, we wondered whether links with adult sleep disorder laboratories could account for some of the differences in CPAP use. Centers 6 and 8, both of which had a high proportion of CPAP use, examined both adults and children with sleep-disordered breathing, whereas center 3 (with a lower proportion of CPAP use) had no links with an adult center. The remaining centers had informal ties with adult centers.

DISCUSSION

The combined experience of a number of major pediatric sleep disorder centers has shown that CPAP is safe and well tolerated by children and adolescents with OSA. The lack of persistent complications of OSA in this population and the small number of patients undergoing tracheostomy indicate that CPAP therapy is efficacious in the pediatric age group.

Continuous positive airway pressure has become available only recently; its first reported use was in 1981.[13] Because of the relatively small population of children with OSA unresponsive to tonsillectomy and adenoidectomy, and the difficulty in obtaining pediatric-sized masks, experience with CPAP use in children was limited until recently. The advent of commercial CPAP equipment, including appropriate pediatric masks, has resulted in a burgeoning use of CPAP in children. However, few data are available regarding use of CPAP in this population. In 1986 the Stanford sleep center reported their experience with CPAP use in 10 children.[5] At that time only three subjects were able to use commercial masks. Treatment of OSA with CPAP was found to be effective, but because of problems with mask leaks or with compliance, only three children could be discharged home with CPAP therapy. Recently the same center reported the use of CPAP in 33 children, with a 60% success rate; failures were due to side effects (epistaxis in one patient), problems with the mask, or poor compliance.[14] To improve compliance, the use of a restraining vest was advocated for young children. Other reports on CPAP therapy for OSA in children have been limited to case reports or abstracts.[6-10] A recent report described the use of CPAP perioperatively in children undergoing adenoidectomy and tonsillectomy for OSA but did not describe long-term use of CPAP.[15]

The combined experience reported in this article is limited by the retrospective nature of the data collection. In addition, because of the variations in diagnostic criteria, polysomnographic techniques and interpretation, and clinical methods among different centers, in-depth analysis and comparison of data were not feasible. Some data, such as compliance with CPAP therapy, were partly subjective. Because only one infant was included in our study, these study results cannot be extrapolated to infants. However, other authors have reported successful use of nasal CPAP in infants with OSA.[16, 17]

The level of airway pressure required in children did not differ from that used in adults.[18] This suggests that the CPAP level is related to the degree of airway obstruction rather than to anatomic or other differences between children and adults, and is consistent with the fact that the upper airway closing pressure is similar in children and adults with OSA.[19] However, a significant number of patients required intermittent increases or decreases in the level of CPAP, as determined by routine reevaluation (i.e., not because of changes in symptoms). Considering that the duration of follow-up of this study was short, it can be anticipated that an even larger number of patients require pressure changes with time. We

therefore recommend that children requiring CPAP therapy undergo periodic polysomnographic assessments of the adequacy of CPAP levels.

The greatest limitation to the use of CPAP in both adults and children is poor compliance. Reasons for poor compliance in adults include inconvenience, discomfort, side effects, claustrophobia, and expense.[11] For pediatric patients both the care giver and the patient must be motivated. In the young or developmentally disabled child, the onus of compliance is on the caregiver. Because the alternative to CPAP in these patients is either tracheostomy or persistent OSA associated with hypoxemia and sleep disruption, the parents are often highly motivated. Self-reporting of CPAP use has been shown to be inaccurate in adult patients. Most pediatric centers used objective measurements of compliance (i.e., the equipment hour meter). However, the hour meter overestimates use if the CPAP machine is allowed to remain on without being attached to the patient. It is possible, therefore, that compliance was lower than was believed by the health care practitioners. Nevertheless, compliance was judged to be adequate in the majority of patients. No center found it necessary to use restraints. The overall success with compliance may be partly due to the use of pediatric health care practitioners, including child psychologists when necessary, and frequent clinical assessments and readings of the CPAP hour meter. The incorporation of low-pressure alarms may further enhance effective CPAP use, by alerting caregivers when the CPAP mask is displaced.

A significant portion of patients in this study had idiopathic OSA persisting after adenoidectomy and tonsillectomy, without other known risk factors for OSA. This suggests that children with OSA should be clinically reevaluated and, if necessary, undergo repeated polysomnography, to identify those who may require CPAP.[20]

We were surprised at the wide variation in CPAP use among the different centers, because all were well-recognized academic centers evaluating large numbers of cases annually. The differences in CPAP use among the different centers suggests the need for further research to define the indications for CPAP therapy. Further study is required to determine whether inpatient or outpatient care is preferable, or whether subgroups of patients would benefit from inpatient therapy; whether it is feasible to institute CPAP during the latter half of the initial polysomnogram or during nap polysomnography, or whether it should be instituted during a second, overnight polysomnogram; and to determine the frequency of reevaluation, whether certain clinical criteria can predict the need for CPAP reevaluation, and whether abbreviated polysomnographic techniques (such as home testing) are sufficient for follow-up evaluation. Further research should also evaluate long-term compliance and efficacy.

We conclude that CPAP is a safe, well-tolerated, and effective therapy for children with OSA, including very young and some developmentally disabled children. Thus CPAP provides an alternative to tracheostomy in patients who do not respond to tonsillectomy and adenoidectomy. We recommend that a trial of CPAP be strongly considered for any child with OSA persisting after tonsillectomy and adenoidectomy, and that a prospective study of CPAP use be performed, so that a consensus may be reached on the treatment of children with persistent OSA.

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