Nasal Continuous Positive Airway Pressure Use In Children With Obstructive Sleep Apnea Younger Than 2 Years of Age*

Ralph Downey III, PhD; Ronald M. Perkin, MD, MA; and Joanne MacQuarrie, RRT, RPSGT

Study objectives: **To assess the efficacy of continuous positive airway pressure (CPAP) in obstructive sleep apnea (OSA) patients who are < 2 years of age.**
Design: **A retrospective chart review of 18 patients from 1992 to 1999 who had OSA confirmed by polysomnography. All patients in this study also completed a separate night of CPAP polysomnography to determine the effectiveness of CPAP in the correction of OSA. Nasal CPAP compliance data were gathered via clinical follow-up examination, telephone interview, or mailed questionnaire.**
Setting: **All patients were studied in the Sleep Disorders Center at Loma Linda University Children's Hospital in Loma Linda, CA.**
Patients: **All patients were < 2 years old.**
Intervention: **After OSA was confirmed by the results of technician-attended nocturnal polysomnography, separate technician-attended nocturnal CPAP polysomnography was completed. On CPAP nights, CPAP pressure was titrated to ameliorate OSA and snoring. CPAP pressure was increased by 2-cm H_2O or 1-cm H_2O increments.**
Results: **Data were analyzed by dependent groups *t* test at $p < 0.05$ level of significance. CPAP statistically improved respiratory parameters significantly when compared to baseline polysomnography. The following four patient subgroups emerged from the analysis: group 1 consisted of six patients who had tracheostomies prior to the CPAP trial, with two patients using CPAP as an alternative to tracheostomy; group 2 consisted of two patients who had previous unsuccessful adenostonsillectomies and who used CPAP successfully, with both having OSA resolution over time; group 3 consisted of four patients who did not tolerate CPAP on the study night; and group 4 consisted of six patients who used CPAP nightly, had OSA resolution over time, and therefore, no longer needed CPAP therapy. Thus, 10 of 18 patients used CPAP either on an interim basis for corrective therapy or as a primary treatment modality for OSA.**
Conclusions: **These data show that children < 2 years of age can tolerate and use CPAP effectively. In several cases, CPAP treatment could be discontinued as OSA resolved over time. The reasons for this are discussed in the text.** ***(CHEST 2000; 117:1608–1612)***

Key words: children; continuous positive airway pressure compliance; infancy; obstructive sleep apnea; nasal continuous positive airway pressure; sleep; sleep disorders

Abbreviations: CPAP = continuous positive airway pressure; OSA = obstructive sleep apnea

The incidence of obstructive sleep apnea (OSA) in children is high, with an estimated proportion between 1% and 3%.[1] The peak incidence of OSA is between ages 2 and 6 years, when lymphoid tissue growth is at its peak.[1] OSA lies on a continuum that ranges from mild snoring to OSA with significant clinical consequences.[2] The effects of OSA are pervasive, including decreased cognitive function,[3] cardiovascular complications such as cor pulmonale, and overt congestive heart failure.[4,5] Moreover, infants < 1 year old with OSA may be at increased risk of sudden infant death syndrome.[6]

Adenotonsillectomy is a common OSA treatment in children between 2 and 6 years of age; however, it is not used as commonly for children < 2 years of

*From the Loma Linda University Medical Center Children's Hospital (Drs. Downey and Perkin, and Ms. MacQuarrie), Loma Linda, CA; and the Jerry L. Pettis Memorial Veterans Administration Medical Center (Dr. Downey), Loma Linda, CA.
Manuscript received July 15, 1999; revision accepted February 8, 2000.
Correspondence to: Ralph Downey III, PhD, Jerry L. Pettis Memorial Veterans Medical Center Sleep Disorders Center (111P), 11201 Benton St, Loma Linda, CA 92357; e-mail: Ralph.Downey@med.va.gov

age. Children with OSA also have a greater risk of respiratory compromise following adenotonsillectomy than children without OSA.[7,8]

Traditionally, while continuous positive airway pressure (CPAP) is the most common OSA treatment in adults, it is not commonly used in children. The reason that CPAP has not been used in this age group in unclear. Perhaps its lack of use is due to a lack of data establishing its utility. CPAP may provide a useful treatment alternative among those for whom surgical correction is not indicated or has failed to resolve the OSA.[9]

Few studies have investigated CPAP use in children < 2 years old. The studies that are available have examined CPAP efficacy in children across a wide age spectrum[9–14] or have focused on those < 1 year old.[13,15,16] Moreover, CPAP is not used in a consistent fashion among even the most experienced centers across a wide age span.[11] One study noted that CPAP use was not routinely or consistently used, even among 11 experienced sleep centers.[15] For example, in one center, CPAP was the primary therapy mode for OSA patients, while in 76% of the centers adenotonsillectomy was the primary therapy mode for OSA. The centers most likely to use CPAP were those closely affiliated with adult sleep centers. No study has focused on children < 2 years of age exclusively.

The purpose of this report is to demonstrate that CPAP is efficacious in children with OSA who are < 2 years of age

Materials and Methods

We retrospectively examined the last 18 patients < 2 years of age who had received a diagnosis of OSA by standard clinical polysomnography and who consented to the use of nasal CPAP as an elective treatment for their OSA. If a correctable cause of OSA was not considered as an effective alternative at the time of evaluation, CPAP was presented to the parents and referring physicians as an alternative treatment for OSA.

All polysomnography was completed using a polysomnograph (model 78D; Grass; Quincy, MA). Measurements included the following: left and right electro-oculogram; EEG (leads C3-A2, O1-A2, and C4-A1); chin electromyogram; airflow (three-bead oral-nasal thermistor); chest wall and abdominal wall movement measurement using inductive plethysmography; end-tidal CO_2 monitoring; one-lead ECG; pulse oximetry with strip recorder (model 3700; Ohmeda; Madison, WI, or model 7000; Novametrix; Wallingford, CT). Pulse wave forms were measured from oximetry and displayed on the polysomnograms to help verify the accuracy of the oximetry readings.

Sleep Scoring

Patient records were scored in accordance with published guidelines. If children had sleep spindles, their sleep architectures were scored in accordance with Rechtshaffen and Kales.[17] If sleep spindles were not noted, sleep was scored as rapid eye movement, non-rapid eye movement, or indeterminate. EEG arousals were scored in accordance with American Sleep Disorders Association established guidelines.[18]

Sleep Respiration

OSA was diagnosed in patients if they had more than one obstructive apnea per hour (no airflow in the presence of chest wall excursion).[11] Obstructive apnea was scored when there was no airflow despite chest wall excursion for at least 6 s, or < 6 s if the obstructive apnea was associated with oxygen desaturation of > 3%. Central apnea was scored in the same manner as obstructive apnea except there was no airflow and no chest wall excursion. Hypopneas were included in the apnea index if there was a > 50% reduction in airflow for > 6 s. The results of each study were considered normal or abnormal in accordance with established norms.[19] Between-scorer agreement for polysomnography was > 90% for sleep and respiratory parameters. End-tidal CO_2 and paradoxical breathing were examined; however, these data were not suitable for reliable analysis.

CPAP Trial Methods

In all study patients, polysomnography established the diagnosis of OSA on a separate baseline polysomnography. On a separate night, CPAP was applied. Only the data from the baseline polysomnography and the first CPAP polysomnography were analyzed for the purpose of this article. Eighteen patients < 2 years of age completed both a baseline polysomnography and a separate night CPAP polysomnography. No patient in this study group under analysis received bilevel positive airway pressure.

Patients did not have training with CPAP before their CPAP study night. Parents had the reasons for CPAP use explained to them and were given information on the known reasons why CPAP was thought to be of benefit for their child. Parents agreed to allow the use of CPAP as a therapy for correcting OSA in their children.

CPAP Titration

The goal of the initial CPAP trial night was to abolish both apnea and snoring while maintaining patient comfort so that the patient and parent would use CPAP at home. CPAP was started at 5 cm H_2O in all patients, and then was titrated by 2.0-cm H_2O increments until both OSA and snoring were abolished. Technicians used their respective judgments and assessments of sleep-EEG arousals, leaks from the masks, and respiratory patterns (apnea) to adjust the CPAP in 1.0-cm H_2O increments to provide the best possible patient comfort. Four patients required more than one CPAP trial night to obtain optimal effectiveness. As the study took place over 7 years, different interfaces have been developed and used. Many of the early patients in this study used custom-made masks, commercially available pediatric-sized masks (Respironics; Murrysville, PA), or nasal pillows. Full-face masks were not needed for the patients in this study.

Home CPAP Use

After the CPAP titration night, parents who agreed to treatment had CPAP available for use in their homes at pressures determined by the results of the CPAP polysomnography. Mask preference on the night of the CPAP study was the mask prescribed for home use. The patient's personal physician, with the sleep center staff, followed each patient. Follow-up for compliance and care was by clinical follow-up and/or questionnaires sent to the patient's home or by telephone conversation to

assess the degree of CPAP use and the reasons for intolerance or discomfort, if any. CPAP timers were not used because not every patient had a CPAP timer on their device. Using this method of compliance checking for some and not for all may have biased our assessment of compliance.

Data Analysis

All data were analyzed using a two-tailed, dependent-groups *t* test, with an *a priori* p level < 0.05 set for statistical significance.

Results

All patients were < 2 years of age at the time of the initial CPAP study; 11 patients were < 1 year of age, and 7 patients were between 1 and 2 years of age.

The studied children had a variety of medical disorders that were thought to be contributing factors to their OSA. Patient diagnoses and final known dispositions are listed in Tables 1 and 2.

Patients selected for CPAP use fell into four group types. In group 1, six OSA patients underwent a tracheostomy to treat their existing OSA. These patients had CPAP applied with their tracheostomies occluded in an attempt to correct their OSA. In this group, four patients required continued tracheostomy and elected not to use CPAP. Two patients used CPAP initially. One patient used CPAP until OSA resolved, and one patient accepted CPAP on the study night, used it for almost 1 year, but then elected tracheostomy as a long-term treatment for OSA. In cases requiring decannulation, an otolaryngologist was consulted.

Group 2 consisted of two patients whose adenotonsillectomies failed to correct their OSA. Both patients had their OSA resolve over time, which allowed CPAP to be discontinued.

Group 3 consisted of four patients who did not accept CPAP on the initial CPAP study night. Of these, one patient was obese with hypothyroidism, two required craniofacial surgery, and one had laryngomalacia and was lost to follow-up (Table 1).

Table 2–*Patients for Whom CPAP Was Used

Patient No./Pre-PSG Diagnosis	Initial CPAP Treatment Pressure, cm H_2O†
1/Laryngomalacia	7; 2 L/min O_2
2/Bronchopulmonary dysplasia	7
3/Laryngomalacia	6; 1 L/min O_2
4/Post-T&A with residual OSA	6; 2 L/min O_2
5/Down's syndrome	7; 2 L/min O_2
6/Laryngomalacia	11
7/CHF	8; 1 L/min O_2
8/Primary OSA, with ALTE history	8
9/Post-T&A with residual OSA	7
10/Pierre Robin syndrome	9

*Patients 1 to 9 received CPAP until resolution of OSA (documented by PSG). Patient 10 received CPAP for 1 year, at which time an elective tracheostomy was performed. ALTE = apparent life-threatening episode; PSG = polysomnography. See Table 1 for other abbreviations.

†Values after the semicolon are for supplemental oxygen therapy.

Group 4 consisted of six patients who received CPAP until follow-up polysomnography demonstrated that their OSA had resolved. These patients had a wide spectrum of disorders, including bronchopulmonary dysplasia, Down's syndrome, laryngomalacia, pharyngeal flap airway obstruction, congestive heart failure, and primary OSA presenting as an apparent life-threatening episode. CPAP was used for approximately 1 to 5 years in this group; three patients had two polysomnography sessions to document the complete resolution of OSA, and three patients had one polysomnography session to document complete resolution.

Table 3 lists the following statistically significant parameters that changed with CPAP treatment: the apnea index was significantly reduced (in all studied patients), as was the time of the longest apnea; and the arterial oxygen saturation nadir improved with CPAP, as did the number of minutes that the patients slept with arterial oxygen saturation at

Table 1–*Patients Who Did Not Use CPAP

Diagnoses	Post-CPAP Trial Disposition
Patients who elected not to use CPAP	
Hypoventilation syndrome	Tracheostomy continued
Hypoventilation, CHF	Tracheostomy continued
Pierre Robin syndrome	Tracheostomy continued
CHF/pulmonary HTN	Tracheostomy continued
Patients intolerant to CPAP on the CPAP trial night	
Pierre Robin syndrome	Craniofacial management
Laryngomalacia	Lost to follow-up
Obesity, hypothyroidism	T&A; UPPP
Crouzon's syndrome	Craniofacial management

*T&A = adenotonsillectomy; UPPP = uvulopalatopharyngoplasty surgery; CHF = congestive heart failure; HTN = hypertension.

Table 3–*Significant Polysomnographic Variables That Changed From Baseline Polysomnogram to CPAP Polysomnogram in Children With OSA (n = 18)***

Variable	PSG		*t* Test (df)
	Baseline	CPAP	
No. of awakenings > 15 s†	13.5 ± 14.8	5.5 ± 5.2	0.9 (15)‡
Apnea index	12.8 ± 20.0	4.5 ± 13.4	4.4 (16)§
Obstructive apnea index	4.7 ± 13.4	2.0 ± 7.3	3.8 (16)§
Hypopnea apnea index	6.7 ± 12.7	2.0 ± 5.7	8.7 (16)‡
Longest apnea, s	25.6 ± 17.4	8.2 ± 7.3	3.9 (17)‖
Minimum Sao_2, %	74.8 ± 20.1	87.3 ± 9.5	2.3 (17)‡
Sao_2 < 90%, min	22.2 ± 25.5	6.4 ± 14.9	2.3 (15)‡

*Values given as mean ± SD. df = degrees of freedom; Sao_2 = arterial oxygen saturation. See Table 1 for other abbreviation.
†For the entire sleep period.
‡$p < 0.05$.
§$p < 0.0001$.
‖$p < 0.001$.

< 90%. Complete abolition of OSA was not always possible due to patient discomfort or severity of the disease.

The mean (SD) arousal index was 16.31 (12.29) while not receiving CPAP and 16.06 (13.56) while receiving CPAP. Five patients needed supplemental oxygen in addition to CPAP (range, 0.5 to 6 L/min) (Table 2). The median CPAP pressure was 7.0 cm H_2O.

Sleep Architecture

Sleep stages did not significantly differ between those measured with baseline polysomnography and CPAP polysomnography. Patients fell asleep in < 13 min, on average, during their baseline studies and with CPAP.

Discussion

These data demonstrate that CPAP is an effective treatment for OSA in patients who are < 2 years old across a wide disease spectrum. This study supports and extends the findings of Guilleminault and colleagues[16] concerning OSA in infants < 1 year of age to those < 2 years of age. An important finding of this study was that OSA resolved over time in nine children. It is unlikely that CPAP "cured" OSA. OSA resolution may have resulted, in part, from the nature of the children's disorders concomitant with OSA, such as bronchopulmonary dysplasia, laryngomalacia, and congestive heart failure. These disorders may resolve with maturity. Moreover, corrective surgery, while not completely successful at correcting OSA in this study group, may have served to widen the upper airway and may have contributed to apnea resolution with maturity. The important point is that CPAP helped these patients safely reach the age at which OSA was resolved.

The success of CPAP in correcting OSA could not be predicted based on the admitting diagnosis. With a small data set, statistical analysis is not likely to reveal a singular predictor of CPAP success. If there is a cautionary tale in this regard, it is that some patients, such as those with Down's syndrome, could easily have been dismissed as "poor candidates for CPAP," but in fact tolerated CPAP well.

A significant number of patients were evaluated after surgery, with six having examinations because of the question of the need for continued tracheostomy. In patients studied with their tracheostomy tube plugged, it was thought that CPAP could not treat their OSA as effectively because there was a question as to whether these patients would tolerate CPAP at home. The subgroups of patients illustrate the heterogeneity of the CPAP population in the young age group studied.

Surprisingly, the EEG arousal index did not change from the baseline polysomnography to the nasal CPAP polysomnography, perhaps due to the lack of CPAP adaptation on the part of the patient. It may be that the arousal index, as a treatment-guiding parameter, may be misleading for a single-night CPAP study. Other indexes of improvement may be more sensitive to change, such as esophageal pressures, which were not measured in this study. If the theory that arousal is due to CPAP is correct, one would expect the arousal index to decrease over time, but that was not examined in this study.

Complications from CPAP in this age group were not unlike those seen in adults, with the most common being mask discomfort, rhinitis, and airway dryness. No patients had pneumothorax or barotrauma.

Further improvements in the use of CPAP in young children may come from studies like this one, which demonstrate that CPAP can be used effectively. With increased support services for CPAP users, CPAP use is likely to increase proportionally with patient and clinician comfort levels. Moreover, with time and the acquisition of more data refining CPAP use in young children, clinicians will find a place for CPAP in the OSA treatment armament.

Conclusion

The main findings of this study were the following: (1) CPAP improved OSA in all study patients; (2) CPAP can be accepted and tolerated well by a majority of patients with severe OSA who are < 2 years of age; (3) CPAP is an effective treatment

modality in children with OSA; and (4) CPAP was not a permanent treatment, as nine patients no longer required CPAP based on follow-up polysomnography-documented resolution of OSA. Some of these patients had self-limiting medical problems for which CPAP may have acted as an interim treatment modality.

A consensus about how CPAP can be used across patient populations in children with OSA would be of benefit to sleep centers in which patients with OSA are frequently seen. This is particularly relevant in instances where little benefit occurs by using surgery or alternate therapies.

ACKNOWLEDGMENTS: The authors thank Michelle Gasca for data compilation and Grennith Zimmerman, PhD, for help in the statistical analysis of the data.

References

1 Rosen C. Obstructive sleep apnea syndrome (OSAS) in children: diagnostic challenges. Sleep 1996; 19(suppl):S274–S277
2 Downey R III, Perkin RM, MacQuarrie J. Upper airway resistance syndrome: sick, symptomatic but underrecognized. Sleep 1993; 16:620–623
3 Dahl RE. The impact of inadequate sleep on children's daytime cognitive function. Semin Pediatr Neurol 1996; 3:44–50
4 Perkin RM, Downey R III, MacQuarrie J. Sleep disordered breathing in infants and children. Respir Care Clin N Am 1999; 5:395–426
5 Brouillette RT, Fernbach SK, Hunt CE. Obstructive sleep apnea in infants and children. J Pediatr 1982; 100:31–40
6 Kahn A, Groswasser J, Rebutffat E, et al. Sleep and cardiorespiratory characteristics of infant victims of sudden death: a prospective case-control study. Sleep 1992; 15:287–292
7 Wiatrak BJ, Myer CMD, Andrews TM. Complications of adenotonsillectomy in children under 3 years of age. Am J Otolaryngol 1991; 12:170–172
8 Williams EFD, Woo P, Miller R, et al. The effects of adenotonsillectomy on growth in young children. Otolaryngol Head Neck Surg 1991; 104:509–516
9 Guilleminault C, Nino-Murcia G, Heldt G, et al. Alternative treatment of tracheostomy in obstructive sleep apnea syndrome: nasal continuous positive airway pressure in young children. Pediatrics 1986; 78:797–802
10 Downey R III, Perkin RM, MacQuarrie J, et al. Six years experience with nasal CPAP in children with obstructive sleep apnea [abstract]. Sleep Res 1997; 26:355
11 Marcus CL, Ward SL, Mallory GB, et al. Use of nasal continuous positive airway pressure as treatment of childhood obstructive sleep apnea. J Pediatr 1995; 127:88–94
12 Waters K, Everett F, Harris MA, et al. Use of nasal mask CPAP instead of tracheostomy for palliative care in two children. J Paediatr Child Health 1994; 30:179–181
13 Waters K, Everett F, Bruderer JW, et al. Obstructive sleep apnea: the use of nasal CPAP in 80 children. Am J Respir Crit Care Med 1995; 152:780–785
14 Brooks L, Sleeper G. Use of nasal CPAP by children in the home [abstract]. Sleep Res 1991; 20:212
15 Marcus CL, Ward SL, Mallory GB, et al. Use of nasal continuous positive airway pressure as treatment of childhood obstructive sleep apnea. J Pediatr 1995; 127:88–94
16 Guilleminault C, Pelayo R, Clerk A, et al, Home nasal continuous positive airway pressure in infants with sleep-disordered breathing. J Pediatr 1995; 127:905–912
17 Rechtschaffen A, Kales A. A manual of standardized terminology, techniques, and scoring system for sleep stages of human subjects. Los Angeles, CA: University of California at Los Angeles Brain Research Institute, 1968
18 American Sleep Disorders Association Sleep Disorders Atlas Task Force. EEG arousals: scoring rules and examples. Sleep 1992; 15:173–184
19 Marcus CL, Omlin KJ, Basinski DJ. Normal polysomnographic values for children and adolescents. Am Rev Respir Dis 1992; 146:1235–1239