NEW RESEARCH

JCSM
Journal of Clinical Sleep Medicine

http://dx.doi.org/10.5664/jcsm.1914

Predictors of Positive Airway Pressure Therapy Adherence in Children: A Prospective Study

Natalie DiFeo[1]; Lisa J. Meltzer, Ph.D.[2]; Suzanne E. Beck, M.D., F.A.A.S.M.[1]; Laurie R. Karamessinis[1]; Mary Anne Cornaglia[1]; Joel Traylor[1]; John Samuel[1]; Paul R. Gallagher, M.A.[3]; Jerilynn Radcliffe, Ph.D.[3]; Heidi Beris, B.S.N.[1]; Mary Kate Menello, B.S.N.[1]; Carole L. Marcus, M.B.B.Ch., F.A.A.S.M.[1]

[1]Sleep Center and [3]Clinical and Translational Research Center, Children's Hospital of Philadelphia, University of Pennsylvania School of Medicine, Philadelphia, PA; [2]Department of Pediatrics, National Jewish Health, Denver, CO

SCIENTIFIC INVESTIGATIONS

Study Objectives: Children with obstructive sleep apnea are increasingly being treated with positive airway pressure (PAP), particularly if they have underlying medical conditions. Although PAP is an effective treatment, its use is challenging due to poor adherence. We hypothesized that demographic, psychosocial, and polysomnographic parameters would be related to PAP adherence. We therefore prospectively collected data potentially pertaining to PAP adherence, and correlated it with PAP use.

Methods: Fifty-six patients and their parents completed a series of psychosocial questionnaires prior to PAP initiation. Objective adherence data were obtained after 1 and 3 months of PAP use.

Results: The population was primarily obese; 23% had neurodevelopmental disabilities. PAP adherence varied widely, with PAP being worn 22 ± 8 nights in month-1, but mean use was only 3 ± 3 h/night. The greatest predictor of use was maternal education ($p = 0.002$ for nights used; $p = 0.033$ for mean h used/night). Adherence was lower in African American children *vs* other races ($p = 0.021$). In the typically developing subgroup, adherence correlated inversely with age. Adherence did not correlate with severity of apnea, pressure levels, or psychosocial parameters other than a correlation between family social support and nights of PAP use in month-3.

Conclusions: PAP adherence in children and adolescents is related primarily to family and demographic factors rather than severity of apnea or measures of psychosocial functioning. Further research is needed to determine the relative contributions of maternal education, socioeconomic status and cultural beliefs to PAP adherence in children, in order to develop better adherence programs.

Keywords: CPAP, obstructive sleep apnea, maternal education

Citation: DiFeo N; Meltzer LJ; Beck SE; Karamessinis LR; Cornaglia MA; Traylor J; Samuel J; Gallagher PR; Radcliffe J; Beris H; Menello MK; Marcus CL. Predictors of positive airway pressure therapy adherence in children: a prospective study. *J Clin Sleep Med* 2012;8(3):279-286.

The obstructive sleep apnea syndrome (OSAS) affects approximately 2% of young children.[1] In most children with OSAS, adenotonsillectomy is the first line of treatment.[1] However, in children with persistent postoperative OSAS due to additional risk factors, such as obesity or genetic syndromes, or in children for whom surgery is contraindicated, treatment with positive airway pressure (PAP) therapy is often required.[1-7] Although PAP use has become common in children, there is a paucity of studies rigorously evaluating its use. In particular, there have been very few studies evaluating PAP adherence in children using objective criteria. Studies in adults have shown that factors that are often thought intuitively by clinicians to be important for PAP adherence, such as high pressures or interface issues,[8,9] have not been borne out by rigorous study.[10]

The few studies available in children suggest that PAP is highly effective in the laboratory situation, but its use outside the laboratory is limited by suboptimal adherence.[3,6] To our knowledge, there have been no prospective studies evaluating psychosocial predictors of PAP use in children. A better understanding of the factors predicting PAP adherence in children would help in formulating better management plans. We therefore prospectively evaluated factors predicting PAP adherence in children referred for PAP initiation. Possible indicators of PAP adherence were chosen based on the literature regarding PAP adherence in adults, and adherence to other medical regimens in children. We hypothesized that older age (i.e., adolescence),[11] minority status,[9,12] and lower maternal education[13,14] would be associated with less adherence. We also hypothesized that low parental stress and higher levels of social support would be associated with increased adherence,[9,13,15] In addition, we evaluated exploratory factors including clinical parameters and parental and child rated quality of life.[16]

This study was conducted prospectively as part of a clinical trial examining PAP adherence in children with OSAS receiv-

BRIEF SUMMARY
Current Knowledge/Study Rationale: Positive airway pressure (PAP) treatment is increasingly being used in children with obstructive sleep apnea, but its efficacy is limited by poor adherence. There have been no prospective studies evaluating predictors of PAP adherence in children.
Study Impact: This study shows that maternal education is the most significant predictor of PAP adherence in children, although race, age and family social support are also predictive factors. These factors should be considered when designing interventions to promote PAP adherence.

Table 1—Survey instruments

Instrument	Description of Measure	Respondent
Modified Epworth Sleepiness Scale[30]	Measure of daytime sleepiness that has previously been adapted for the pediatric population[30-32]	Caregiver
Child Behavior Checklist (CBCL)[33,34]	Survey of behavior competencies that yields standardized, age-adjusted scores on internalizing and externalizing behavior difficulties	Caregiver (all) plus youth self-report if 11-18 years of age and developmentally able
Conners Abbreviated Symptom Questionnaire[35]	Screening tool for attention deficit hyperactivity disorder	Caregiver
Pediatric Quality of Life (PedQL)[36]	A standard measure of quality of life that has been used widely in pediatric populations	Caregiver (all) plus child if ≥ 5 years of age and developmentally able
OSA18[37]	An instrument that combines symptoms of OSAS and disease-specific quality of life measures	Caregiver
Parenting Stress Index Short Form (PSI-F)[38]	A measure of parents' perception of stress in the parent-child relationship	Caregiver
Medical Outcomes Study Social Support (MOSS)[39]	A measure of perceived social, emotional, and tangible parental support	Caregiver
Nasal Obstruction Symptom Evaluation (NOSE)[40]	A measure of nasal symptoms	Caregiver with child input

ing two modes of PAP delivery: continuous positive airway pressure (CPAP) *vs* bilevel pressure release (Bi-Flex). Although no difference in adherence was found between modes,[17] the mode of usage was controlled for in the current study. The study was registered at Clinicaltrials.gov (#NCT00458406).

METHODS

Study Population

Consecutive children aged 2-16 years with polysomnographically diagnosed OSAS, who were naïve to PAP and who were referred for PAP initiation, were recruited from the Sleep Center at The Children's Hospital of Philadelphia. The study was approved by the Children's Hospital of Philadelphia Institutional Review Board (#07-005190). Written informed consent was obtained from the parent/legal guardian; and assent from children ≥ 7 years of age when capable.

Measures

All subjects had undergone a baseline clinical polysomnogram prior to study entry, and medical records were reviewed. Height and weight were measured, and subjects were classified as obese if their body mass index was ≥ 95th percentile for age and sex.[18] Subjects and the accompanying parent/guardian completed a series of psychosocial questionnaires (see details of the measures in **Table 1**).

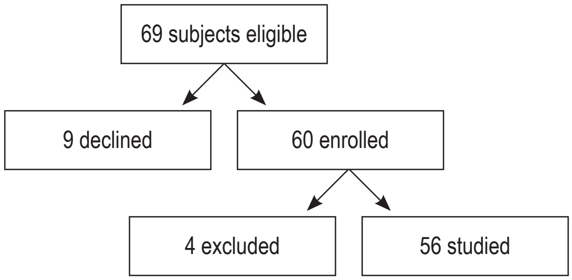
PAP Protocol

Subjects were fitted with a PAP mask and allowed to habituate to this for 2 weeks. They then underwent a protocol-driven titration polysomnogram. Subjects received free PAP equipment, including heated humidification and interfaces, the morning after titration. A 48-h phone call, 2-week phone call and monthly visits to the research clinic were made to assess use, provide support, and identify and troubleshoot side effects and barriers to use, such as behavioral issues, nasal congestion, eye irritation, skin breakdown, or equipment problems. The type of interface was adjusted or changed as needed. All efforts were made to contact subjects who did not use their PAP or return for evaluations and to address and treat any potential barriers to PAP use, e.g., by using behavior modification techniques and treating nasal symptoms with antihistamines, decongestants, or anti-inflammatories. Objective adherence data were obtained from the equipment at month-1 and month-3 (EncorePro2, Respironics, Murrysville, PA). Primary outcomes were the mean number of nights the equipment was placed on per month, and mean hours of PAP use at prescribed pressure per night (averaged over all nights). In addition, the number of hours that the machine was turned on but did not necessarily deliver the prescribed pressure was measured at month-1.

Statistical Methods

Unless otherwise specified, data are shown as mean ± SD. A p-value < 0.05 was considered significant. Histograms and Kolmogorov-Smirnov tests were used to examine normalcy of distribution. Appropriate parametric or nonparametric tests were used to examine differences between subjects under 2 conditions as well as differences between subgroups, and differences between a variety of categorical variables, including obesity status (Yes/No), gender, race (African American *vs* all others), and developmental delay (Yes/No). Maternal education was treated as an ordered categorical variable. Appropriate correlation coefficients (Pearson or Spearman) between predictor variables and PAP were examined. Hierarchical multiple linear regression models were used to examine the effects of various covariates on the primary PAP use outcomes. Variables correlating with PAP use at the < 0.10 significance level and variables considered theoretically important were considered for inclusion in the pool of potential predictors, and were entered in separate blocks in a specified order. PAP mode was forced into the model in the first block. Developmental delay

Figure 1—Flow diagram of subject enrollment



Sixty-nine consecutive children were approached, of whom 9 declined (2 because of distance, 5 because of the burden of the study visits, 1 for no stated reason, and 1 because of severe parental illness). Thus, 60 children were enrolled. Four children were subsequently excluded: one child with Down syndrome developed pneumonia one week after enrollment and underwent tracheotomy; one child with nasopharyngeal stenosis underwent surgery; one adolescent was institutionalized for behavioral problems without his PAP equipment; and one child moved away after consenting but before entering the study. Thus, data are presented on 56 subjects.

(Yes/No) and CBCL score (disruptive child behaviors) were entered in the second block; total scores of the PSI (parental stress) and MOSS (social support) were entered in the third block; and age, race, and maternal education were entered in blocks 4-6, respectively. At block 2 through 6, a stepwise entry method was used.

RESULTS

Study Group

Sixty children were enrolled; 4 were subsequently excluded. See **Figure 1** for details of enrollment and exclusion. Adherence data could not be obtained for 2 patients at month-1 due to equipment malfunction, but could be obtained at month-3. Two subjects were evaluated at month-1 but were subsequently lost to follow-up.

Subject characteristics are shown in **Table 2**. As is typical for children requiring CPAP,[3,5,6] many subjects were obese and/or had underlying medical problems contributing to their OSAS, and almost a quarter had major developmental disabilities. Children had moderate to severe OSAS and excessive daytime sleepiness.

Table 2—Study group

N	56
Age (yr)	12 ± 4
Range	2–16
Males	38 (68)
Race	
African American	33 (59)
Caucasian	20 (36)
More than one race	3 (5)
Hispanic ethnicity	5 (9)
Obese[a]	40 (71)
Other diagnoses[a]	
Genetic syndrome	11 (20)
Central nervous system abnormality	6 (11)
Craniofacial syndrome	3 (5)
Pulmonary disease	3 (5)
Growth hormone deficiency	1 (2)
Neurodevelopmental disability[a]	13 (23)
Maternal education	
Did not complete high school	2 (4)
Completed high school; no college	16 (29)
Some college	21 (38)
Completed college	10 (18)
Completed postgraduate degree	7 (13)
Polysomnographic results	
Apnea hypopnea index (N/h)	19 ± 16
Arterial oxygen saturation nadir (%)	79 ± 13
Peak end-tidal CO_2 (mm Hg)[b]	57 ± 5
Modified Epworth score[30]	10 ± 5
PAP pressure	
Subjects on CPAP (N = 13)	
CPAP (cm H_2O)	8 ± 2
Subjects on bilevel pressure (N = 43)	
Inspiratory pressure (cm H_2O)	13 ± 3
Expiratory pressure (cm H_2O)	7 ± 2

Data shown as mean ± SD or N (%) unless otherwise specified. [a]Note that some children had multiple diagnoses. [b]N = 52; not obtained in 4 subjects who had their baseline sleep study at outside facilities.

Adherence

Subjects placed PAP on most nights, with a mean use of 22 ± 8 nights in month-1 (**Figure 2**); 78% used it ≥ 50% of nights for month-1. At month-3, mean use was slightly lower (19 ± 9 nights/month). However, although PAP was placed on the subject most nights, the mean duration of nightly use was low. PAP was used for only 3 ± 3 h/night (range 0-9) during month-1, although there was wide individual variation. Month-3 data were similar, with mean nightly use of 2.8 ± 2.7 hours.

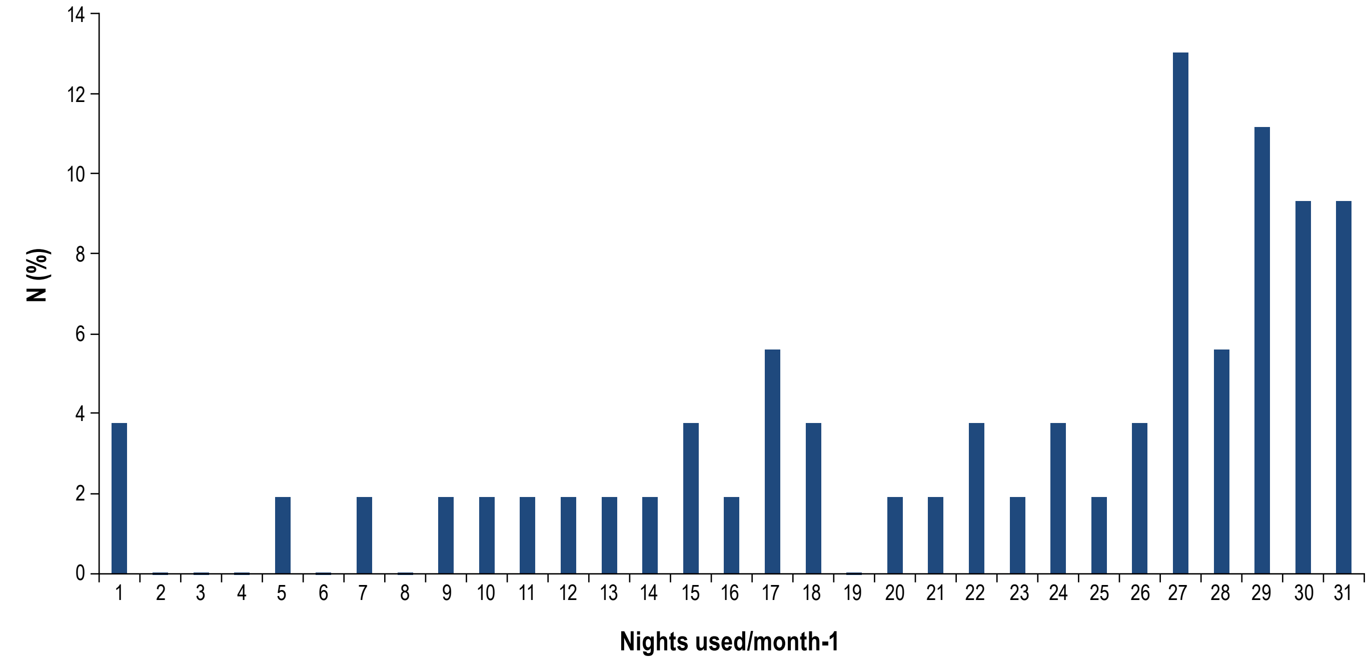
The average number of h/night during month-1 that the device was turned on (i.e., the total time that the device was turned on, whether it was delivering full pressure or not) was much higher than the mean use at pressure: 4.9 ± 2.7 *vs* 3 ± 3 h ($p < 0.001$).

Factors Predicting PAP Adherence

There was no correlation between adherence and age for the group as a whole (**Figure 3**). When the subjects with developmental delay were excluded from analysis, there was a significant inverse correlation between age and number of nights at month-3 ($r = -0.483$, $p = 0.001$), but not for other adherence parameters.

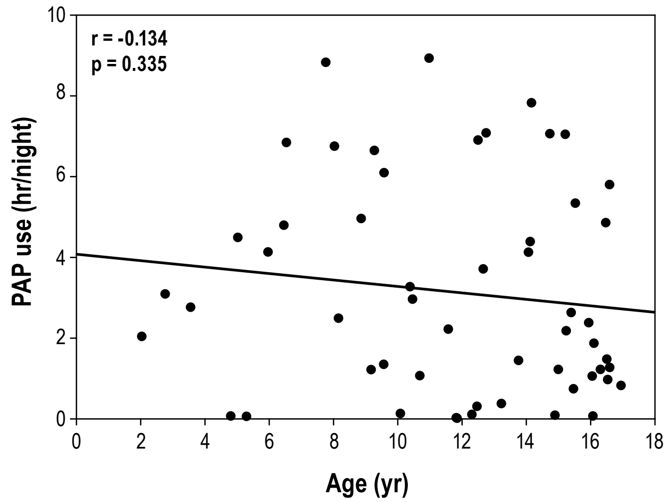
African American children used PAP for fewer hours/night than other races (2.5 ± 2.4 *vs* 4.2 ± 2.8 h in month-1, $p = 0.021$). However, there was no significant difference in the number of nights that PAP was placed on (African Americans 21.6 ± 8.6 *vs* other races 23.2 ± 8.0 nights, $p = 0.63$), or in the number

Figure 2—Positive airway pressure nightly use for month-1



The number of nights/month that positive airway pressure therapy was used during month-1 is shown for individual subjects (expressed as a percentage of total subjects)

Figure 3—Correlation between positive airway pressure use and age for month-1



The correlation between mean nightly positive airway pressure (PAP) use and age is shown for month-1. There was no significant correlation.

of hours of use/night for month-3. There were no differences between African American subjects and subjects of other races in any of the demographic, polysomnographic, behavioral, or social predictor variables, with the exception of developmental delay, which was less common in the African American subjects ($p = 0.026$).

Adherence was highly correlated with maternal education (**Figures 4**, **5**): Month-1, $r = 0.405$, $p = 0.002$ for nights used, and $r = 0.290$, $p = 0.033$ for mean h of use/night; month-3, $r = 0.396$, $p = 0.004$ for nights used, and $r = 0.314$, $p = 0.023$ for mean h of use/night. Note that maternal education was used, as only 9% of respondents were fathers.

There was no effect on adherence of gender, obesity status, presence/absence of developmental delay, severity of baseline polysomnography, pressure level, nasal symptoms at baseline, sleepiness, child behavior, parental stress, or social support, except for the MOSS, which correlated with nights used at month-3 (i.e., a higher level of social support correlated with more PAP use; $r = 0.50$, $p < 0.0005$). Parental stress, as measured by the Parental Stress Index, did not change between month-1 and month-3 ($p = 0.47$).

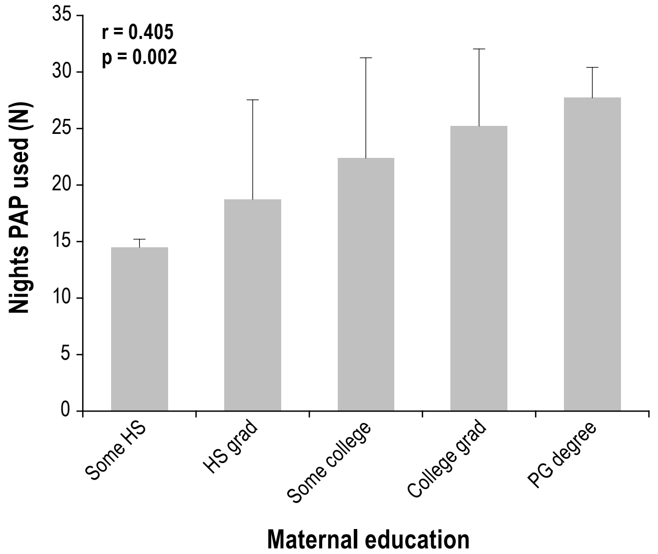
Other than the effect of age (noted above), findings were similar when the subset of children with developmental delay was excluded from analysis.

PAP has not been approved by the Food and Drug Administration (FDA) in the USA for children < 7 years of age or weighing < 40 pounds (18.2 kg). Nine of the subjects were < 7 years of age, and 4 of these children also weighed < 18.2 kg and therefore did not meet FDA eligibility criteria. There was no difference in PAP adherence between children < 7 years of age compared to older children.

Regression Models

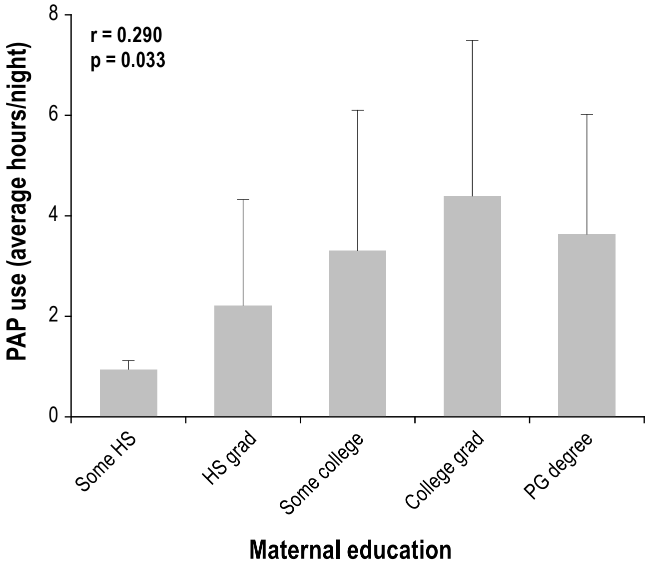
Data from the regression models are shown in **Table 3**. The unstandardized regression coefficients (β) indicate that for a unit increase in maternal education (e.g., from high school graduate to some college), ~3.5 more nights of PAP use would be expected at month-1; that the comparison group (race other than African American) would be expected to use PAP ~1.9 h

Figure 4—Maternal education and number of nights with positive airway pressure use (month-1)



The number of nights that positive airway pressure (PAP) was used at each level of maternal education is shown for month-1. There was a significant correlation between maternal education and nights PAP was used. HS, high school; grad, graduate; PG, postgraduate.

Figure 5—Maternal education and hours of PAP use per night (month-1)



The mean hours per night of positive airway pressure (PAP) use per night at each level of maternal education is shown for month-1. There was a significant correlation between maternal education and minutes of PAP use. HS, high school; grad, graduate; PG, postgraduate.

Table 3—Multiple linear regression model results, controlling for positive airway pressure mode (mode forced into the model at step 1)

Independent variable	Unstandardized β coefficient	SE	β coefficient p value	Change in R^2	p-value for change in R^2	Overall R^2	Overall p value
Outcome = Nights Used, Month 1							
Constant	-3.157	7.507	0.676			–	–
PAP mode	1.859	2.479	0.457	0.006	0.585	–	–
Maternal education	3.480	1.026	0.001	0.197	0.001	0.203	0.005
Outcome = Nights Used, Month 3							
Constant	-4.367	6.289	0.491			–	–
PAP mode	-1.412	2.690	0.602	0.006	0.597	–	–
MOSS	5.466	1.399	< 0.0005	0.252	< 0.0005	0.258	0.001
Outcome = Mean Nightly Use (hours/night), Month 1							
Constant	2.442	0.490	< 0.0005			–	–
PAP mode	-0.066	0.840	0.938	0.001	0.831	–	–
Race[a]	1.943	0.732	0.011	0.130	0.011	0.131	0.037
Outcome = Mean Nightly Use (hours/night), Month 3							
Constant	2.707	0.470	< 0.0005			–	–
PAP mode	-1.229	0.870	0.165	0.030	0.240	–	–
Developmental delay[b]	2.401	0.927	0.013	0.126	0.013	0.156	0.022

The unstandardized regression coefficient (β), standard error (SE) of the coefficient, p value of the coefficient, change in R^2 and its p-value as a result of the addition of the new predictor, overall R^2 for the entire model and overall p value for the model are shown for each adherence outcome. Note that the unstandardized regression coefficient (β) reflects the change in the outcome per unit change in the predictor variable. PAP, positive airway pressure. [a]Coded 0 = African American, 1 = other. [b]Coded 0 = No, 1 = Yes.

longer per night at month-1 compared to the reference group (African American); and that the subjects with developmental delays would be expected to use PAP ~2.4 h longer per night at month-3 compared to typically developing children. Changes in R^2 at each step as a result of adding the new predictor at that step are shown.

DISCUSSION

This study evaluated a wide range of demographic, psychosocial, and polysomnographic variables that were thought to be possible predictors of PAP adherence in children and adolescents. Despite the removal of any potential economic barrier by providing free equipment and treatment, it was shown that lower maternal education was the strongest predictor of poor PAP adherence, with older, typically developing youth, and African American youth less adherent to PAP. Lower levels of social support were also associated with poor adherence.

Similar to other studies of pediatric PAP use, this study comprised a clinically heterogeneous group of children with many medical and psychiatric comorbidities.[2-7] Adults with OSAS requiring PAP treatment are typically obese but otherwise healthy. In contrast, most children with OSAS respond clinically to adenotonsillectomy, and only a small proportion go on to require PAP[1]; thus, those requiring PAP are typically children with additional medical risk factors for OSAS such as Down syndrome. Therefore, PAP adherence programs need to be designed for medically complex and/or developmentally delayed children and their families. The proportion of obese and African American children in this study reflects an inner city population which may not be representative of other areas, although the increasing prevalence of childhood obesity has resulted in a worldwide increase in obesity and childhood OSAS.[19,20]

As found in other studies of adherence to pediatric medical regimens,[13] maternal education was strongly associated with adherence to PAP. Presumably, this was due to better parental understanding of the consequences of OSAS and the importance of treatment; in general, parental literacy and education have been associated with better child health outcomes, including better adherence to medical regimens.[21] However, it is also possible that the relationship between PAP adherence and maternal education was a reflection of socioeconomic status. Low socioeconomic status has been associated with poor adherence to CPAP in adults,[9] and poor adherence to other medical regimens in children.[22] A limitation of this study was the lack of data regarding socioeconomic status.

After excluding the subgroup of children with developmental disabilities, older children and adolescents were less likely to be adherent to PAP than younger children. This was not surprising, as adolescents tend to have poor adherence to medical regimens,[11] and a study of subjective PAP use in adolescents showed poor adherence.[23] The poor adherence in adolescents may be related to developmental issues such as feeling invincible, not wanting to be different from peers, not perceiving potential benefits from therapy, and the overall need to rebel against authority. In addition, it is likely that the older children and adolescents had less parental supervision for PAP use. A recent study of children with asthma found that 50% of 11-year-olds and 75% of 15-year-olds were responsible for taking their own medication[24]; it is likely that the adolescents in the current study had similar responsibilities for their PAP use. Indeed, a study of adolescents with diabetes showed that adherence improved when family members were more involved in the patient's care.[13] This may be one reason why children with developmental disabilities, whose parents are more closely involved in their care, did not show age-related differences in adherence.

African American children were less likely to be adherent than those of other races. This is similar to studies of adults, in whom African Americans were five times more likely to be nonadherent to PAP than Caucasians.[9] It is possible that racial differences in adherence are due to differences in socioeconomic status and maternal education level, but differences may also be related to cultural attitudes and beliefs.

Similar to other studies of adherence in children[13] and adults,[9,15] indices of family social support were somewhat predictive of PAP adherence. Concordant with studies in adults, neither polysomnographic measures of OSAS severity nor nasal symptoms predicted PAP adherence.[9] Daytime sleepiness, a predictor of PAP adherence in adults,[25] was not a predictor of adherence in this study, perhaps because sleepiness is a less prominent symptom in children with OSAS than in adults.[26] Behavioral factors from the Conners scale and CBCL, such as hyperactivity and externalizing symptoms (e.g., aggression), which are often associated with childhood OSAS,[27] were also not related to PAP adherence.

Adherence decreased slightly between month-1 and month-3, and different factors played a role at each time point (**Table 3**). There was no indication that the clinical or social status of any of the subjects or their families changed during the short observation period. This suggests that factors affecting initiation of PAP are different than factors maintaining PAP use, and future studies should evaluate predictors of long-term PAP use.

A limitation of this study is that self-efficacy, a predictor of PAP adherence in adults,[28] was not studied as we could not find an appropriate measure to use in children. Future studies of self-efficacy in children are needed.

Overall, PAP adherence in this study was suboptimal. Few studies have evaluated PAP adherence in children using objective recordings of PAP use. Two recent studies both showed that about a third of children either refused CPAP immediately or were lost to follow-up; for those who did use CPAP, nightly usage was 5 hours/night.[3,6] However, these data cannot be compared directly to the data in the current paper, as the hourly use in the previously reported studies included only those subjects who accepted CPAP. In contrast, the current study included all children regardless of CPAP use and acceptance, and collected adherence data on 96% of the cohort by month-3. To our knowledge, this is the first study prospectively evaluating predictors of PAP use in children without excluding any who initially refused CPAP. Like other studies using objective adherence data, use was found to be less than reported in studies with subjective reports of CPAP use.[5]

PAP has not been approved by the FDA for use in young children. However, we did not detect any differences in adherence in children < 7 years of age compared to older children, and none of the young children experienced any serious complications from PAP. Home PAP has been used extensively off-label in children ranging in age from infancy onwards, with more than 300 children enrolled in several large published studies.[2-6] It has been shown to be safe and efficacious, especially when compared to the alternative treatments (such as tracheostomy) available for children who are not candidates for or were not cured by adenotonsillectomy.

In summary, this study has shown that key barriers to PAP adherence in children include low maternal education, African American race, older patient age, and decreased family social support, with maternal education being the most prominent predictive factor. Clinical factors such as PAP pressure and nasal symptoms do not predict PAP usage. Pediatric PAP adherence programs tend to focus on equipment modification, behavioral modification and intensive family counseling.[5,6,29] However, this may not be adequate as these programs may not be sufficiently tailored to the needs of many families being served, i.e., families of different races, low socioeconomic status and/or limited maternal education. Further research is needed to determine the relative contributions of maternal education, race, socioeconomic status and cultural and personal beliefs to PAP adherence, and whether a more targeted and culturally tailored focus on patient and family education regarding OSAS and its treatment can improve PAP adherence, and hence the outcomes of children with OSAS.

ABBREVIATIONS

CBCL, Child Behavior Checklist
CPAP, continuous positive airway pressure
FDA, Food and Drug Administration
MOSS, Medical Outcomes Study Social Support
NOSE, Nasal Obstruction Symptom Evaluation
OSAS, obstructive sleep apnea syndrome
PAP, positive airway pressure
PedQL, Pediatric Quality of Life
PSI-F, Parenting Stress Index Short Form

REFERENCES

1. American Academy of Pediatrics. Clinical practice guideline: diagnosis and management of childhood obstructive sleep apnea syndrome. *Pediatrics* 2002;109:704-12.
2. Waters KA, Everett FM, Bruderer JW, Sullivan CE. Obstructive sleep apnea: The use of nasal CPAP in 80 children. *Am J Respir Crit Care Med* 1995;152:780-5.
3. Marcus CL, Rosen G, Ward SL, et al. Adherence to and effectiveness of positive airway pressure therapy in children with obstructive sleep apnea. *Pediatrics* 2006;117:e442-e451.
4. Guilleminault C, Pelayo R, Clerk A, Leger D, Bocian RC. Home nasal continuous positive airway pressure in infants with sleep- disordered breathing. *J Pediatr* 1995;127:905-12.
5. Uong EC, Epperson M, Bathon SA, Jeffe DB. Adherence to nasal positive airway pressure therapy among school-aged children and adolescents with obstructive sleep apnea syndrome. *Pediatrics* 2007;120:e1203-e1211.
6. O'Donnell AR, Bjornson CL, Bohn SG, Kirk VG. Compliance rates in children using noninvasive continuous positive airway pressure. *Sleep* 2006;29:651-8.
7. Massa F, Gonsalez S, Laverty A, Wallis C, Lane R. The use of nasal continuous positive airway pressure to treat obstructive sleep apnoea. *Arch Dis Child* 2002;87:438-43.
8. Ryan S, Garvey JF, Swan V, Behan R, McNicholas WT. Nasal pillows as an alternative interface in patients with obstructive sleep apnoea syndrome initiating continuous positive airway pressure therapy. *J Sleep Res* 2011;20:367-73.
9. Weaver TE, Grunstein RR. Adherence to continuous positive airway pressure therapy: the challenge to effective treatment. *Proc Am Thorac Soc* 2008;5:173-8.
10. Aloia MS. Understanding the problem of poor CPAP adherence. *Sleep Med Rev* 2011;15:341-2.
11. Smith BA, Shuchman M. Problem of nonadherence in chronically ill adolescents: strategies for assessment and intervention. *Curr Opin Pediatr* 2005;17:613-8.
12. Drotar D, Bonner MS. Influences on adherence to pediatric asthma treatment: a review of correlates and predictors. *J Dev Behav Pediatr* 2009;30:574-82.
13. Hsin O, La Greca AM, Valenzuela J, Moine CT, Delamater A. Adherence and glycemic control among Hispanic youth with type 1 diabetes: role of family involvement and acculturation. *J Pediatr Psychol* 2010;35:156-66.
14. DeWalt DA, Hink A. Health literacy and child health outcomes: a systematic review of the literature. *Pediatrics* 2009;124 Suppl 3:S265-S274.
15. Lewis KE, Seale L, Bartle IE, Watkins AJ, Ebden P. Early predictors of CPAP use for the treatment of obstructive sleep apnea. *Sleep* 2004;27:134-8.
16. Modi AC, Quittner AL. Barriers to treatment adherence for children with cystic fibrosis and asthma: what gets in the way? *J Pediatr Psychol* 2006;31:846-58.
17. Marcus CL, Beck SE, Traylor J, et al. Randomized, double-blind clinical trial of two different modes of positive airway pressure therapy on adherence and efficacy in children. *J Clin Sleep Med* 2012;8:37-42.
18. Rosner B, Prineas R, Loggie J, Daniels SR. Percentiles for body mass index in U.S. children 5 to 17 years of age. *J Pediatr* 1998;132:211-22.
19. Bhattacharjee R, Kheirandish-Gozal L, Spruyt K, et al. Adenotonsillectomy outcomes in treatment of obstructive sleep apnea in children: a multicenter retrospective study. *Am J Respir Crit Care Med* 2010;182:676-83.
20. Li AM, So HK, Au CT, et al. Epidemiology of obstructive sleep apnoea syndrome in Chinese children: a two-phase community study. *Thorax* 2010;65:991-7.
21. DeWalt DA, Hink A. Health literacy and child health outcomes: a systematic review of the literature. *Pediatrics* 2009;124 Suppl 3:S265-74.
22. Vreeman RC, Wiehe SE, Pearce EC, Nyandiko WM. A systematic review of pediatric adherence to antiretroviral therapy in low- and middle-income countries. *Pediatr Infect Dis J* 2008;27:686-91.
23. Beebe DW, Byars KC. Adolescents with obstructive sleep apnea adhere poorly to positive airway pressure (PAP), but PAP users show improved attention and school performance. *PLoS One* 2011;6:e16924.
24. Orrell-Valente JK, Jarlsberg LG, Hill LG, Cabana MD. At what age do children start taking daily asthma medicines on their own? *Pediatrics* 2008;122:e1186-e1192.
25. Olsen S, Smith S, Oei T, Douglas J. Health belief model predicts adherence to CPAP before experience with CPAP. *Eur Respir J* 2008;32:710-7.
26. Gozal D, Wang M, Pope DW. Objective sleepiness measures in pediatric obstructive sleep apnea. *Pediatrics* 2001;108:693-7.
27. Beebe DW. Neurobehavioral morbidity associated with disordered breathing during sleep in children: a comprehensive review. *Sleep* 2006;29:1115-34.
28. Aloia MS, Arnedt JT, Stepnowsky C, Hecht J, Borrelli B. Predicting treatment adherence in obstructive sleep apnea using principles of behavior change. *J Clin Sleep Med* 2005;1:346-53.
29. Koontz KL, Slifer KJ, Cataldo MD, Marcus CL. Improving pediatric compliance with positive airway pressure therapy: the impact of behavioral intervention. *Sleep* 2003;26:1010-5.
30. Melendres MC, Lutz JM, Rubin ED, Marcus CL. Daytime sleepiness and hyperactivity in children with suspected sleep-disordered breathing. *Pediatrics* 2004;114:768-75.
31. Elkhayat HA, Hassanein SM, Tomoum HY, Abd-Elhamid IA, Asaad T, Elwakkad AS. Melatonin and sleep-related problems in children with intractable epilepsy. *Pediatr Neurol* 2010;42:249-54.
32. Chan EY, Ng DK, Chan CH, et al. Modified Epworth Sleepiness Scale in Chinese children with obstructive sleep apnea: a retrospective study. *Sleep Breath* 2009;13:59-63.
33. Achenbach TM, Rescorla LA. *Manual for the ASEBA school-age forms & profiles*. Burlington, VT: University of Vermont , Research Center for Children, Youth & Families, 2001.
34. Achenbach TM, Rescorla LA. *Manual for the ASEBA preschool forms & profiles*. Burlington, VT: University of Vermont, Research Center for Children, Youth & Families, 2000.
35. Conners CK, Sitarenios G, Parker JD, Epstein JN. The revised Conners' Parent Rating Scale (CPRS-R): factor structure, reliability, and criterion validity. *J Abnorm Child Psychol* 1998;26:257-68.
36. Varni JW, Seid M, Kurtin PS. PedsQL 4.0: reliability and validity of the Pediatric Quality of Life Inventory version 4.0 generic core scales in healthy and patient populations. *Med Care* 2001;39:800-12.
37. Franco RA Jr., Rosenfeld RM, Rao M. First place--resident clinical science award 1999. Quality of life for children with obstructive sleep apnea. *Otolaryngol Head Neck Surg* 2000;123:9-16.
38. Abidin RR. Parenting Stress Index. Lutz,FL: Psychological Assessment resources, Inc., 1995.
39. Sherbourne CD, Stewart AL. The MOS social support survey. *Soc Sci Med* 1991;32:705-14.
40. Stewart MG, Witsell DL, Smith TL, Weaver EM, Yueh B, Hannley MT. Development and validation of the Nasal Obstruction Symptom Evaluation (NOSE) scale. *Otolaryngol Head Neck Surg* 2004;130:157-63.

ACKNOWLEDGMENTS

The authors thank the patients and their families who participated in this study, all the sleep technologists at the Sleep Center of Children's Hospital of Philadelphia for their help, and Kelly Ann Davis for assistance with data entry.

SUBMISSION & CORRESPONDENCE INFORMATION

Submitted for publication November, 2011
Submitted in final revised form January, 2012
Accepted for publication January, 2012

Address correspondence to: Carole L. Marcus, M.B.B.Ch., Children's Hospital of Philadelphia, Main Building, 9NW50, 34th Street & Civic Center Boulevard, Philadelphia, PA 19104; Tel: (267) 426-5842; Fax: (267) 426-9234; E-mail: marcus@email.chop.edu

DISCLOSURE STATEMENT

This was an investigator-initiated (CLM) study funded by Philips Respironics. All data collection, statistical analyses and manuscript writing were performed by the investigators independent of Philips Respironics. Dr. Marcus also received support from NIH RO1 HL58585, and Dr. Meltzer from K23 MH077662. Off-label use: This study used positive airway pressure therapy in children under 7 years of age or 40 lb.