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Improving Positive Airway Pressure Adherence in Children

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INTRODUCTION

Obstructive sleep apnea syndrome (OSAS) is estimated to affect approximately 1% to 5% of children between the ages of 5 and 13 years.^{1–4} OSAS has been shown to contribute to neurobehavioral and cognitive morbidity in children, including poor school performance, impaired language skills, inattention, hyperactivity, and reduced quality of life.^{5–10} OSAS is also associated with significant physiologic consequences, including insulin resistance, elevated markers of inflammation, systemic hypertension, and, in severe cases, left ventricular dysfunction and cor pulmonale.^{11–17}

Pathophysiologic mechanisms for the development of OSAS can be broadly divided into anatomic factors that reduce the caliber of the upper airway, such as craniofacial abnormalities, obesity, and adenotonsillar hypertrophy, and factors that increase upper-airway collapsibility, such as neurologically based alterations in upper airway muscle tone.^{18–20} Deficits in central control of ventilation (eg, Chiari malformation) may also play an etiologic role in some cases.

INDICATIONS FOR POSITIVE AIRWAY PRESSURE

The primary risk factors for OSAS in children are distinct from those in the adult population, with adenotonsillar hypertrophy being implicated in most childhood cases.^{21–23} Adenotonsillectomy is therefore considered to be the first-line treatment for otherwise healthy children with OSAS.¹ A recent large, randomized controlled trial demonstrated the efficacy of early adenotonsillectomy in improving behavior, quality of life, and polysomnographic features of childhood OSAS.¹⁰ Although most patients with OSAS will undergo surgery, not all pediatric patients with OSAS are appropriate surgical candidates. In addition, some patients continue to be symptomatic and have polysomnographic evidence of

residual OSAS after adenotonsillectomy; risk factors include severe preoperative OSAS, obesity, neuromuscular disorders characterized by hypotonia, and craniofacial abnormalities.^{24–26} These patients may be candidates for positive airway pressure (PAP) therapy.²⁷

MECHANISM OF ACTION OF PAP

PAP is a noninvasive method of treating OSAS, and is the most common treatment modality in adults. The basic mechanism involves it delivering intraluminal airway pressure that is above the critical closing pressure of the airway to overcome dynamic obstruction by stenting the airway open.^{28,29} This positive pressure is generated by an air compressor in the continuous PAP (CPAP) machine, and is transmitted to the patient's airway through a single conduit that connects with the patient via a variety of interfaces, including nasal pillows, nasal masks (Fig. 1), and full-face masks. The device continually adjusts its output to the patient's breathing pattern to maintain a constant pressure. A fixed leak valve is positioned near the mask to prevent rebreathing of carbon dioxide. Fig. 1 shows a typical PAP apparatus.

It must be noted that it is not the movement of air but rather the pressure generated and maintained through the airway that prevents obstructive events. Selection of an appropriately sized mask that provides a snug fit devoid of air leaks is central to maintenance of positive pressure throughout the system. Mask straps, cushions around the interface, and forehead spacers facilitate mask fit and comfort.

Contemporary CPAP devices have evolved in both size and function, with devices becoming more portable and technologically advanced. Most CPAP machines available today are equipped with data-storage capabilities, digitally recording the number of apnea events and air leaks over time. Data can be downloaded from the machine during follow-up, and serve as an objective measure of CPAP usage to provide feedback to the patient and family to aid in improving adherence. In addition, the incorporation of a humidifier in PAP devices has been demonstrated in adults to decrease drying of the nasal mucosa that can lead to nasal congestion, dryness, and epistaxis, and to increase adherence.^{30,31}

EFFICACY OF PAP FOR THE TREATMENT OF CHILDHOOD OSAS

Although it should be noted that the use of home CPAP for children weighing less than 40 lb (18 kg) or younger than 7 years has not been approved by the US Food and Drug Administration, extensive clinical experience and numerous studies have demonstrated CPAP to be safe and efficacious in children of all ages, including infants.^{32–34} Marcus and colleagues²⁷ demonstrated PAP therapy (both CPAP and bilevel PAP [BPAP]) to be effective in improving respiratory parameters in pediatric patients aged 2 to 16 years with OSAS, with highly significant improvements in both the apneahypopnea index (AHI; number of apneas and hypopneas per hour of sleep) ($P = .003$) and oxyhemoglobin saturation nadir ($P = .001$) 6 months after initiating PAP therapy.²⁷ These results were accompanied by significant improvements in subjective parental assessment of sleepiness, snoring, and difficulty breathing during sleep.

CHALLENGES OF PAP ADHERENCE

Benefit from CPAP therapy is predicated on a snugly fit mask that can maintain positive pressure to stent the upper airway. Adherence with the device can be difficult, especially for children with developmental delays, anxiety, or other behavioral problems. These children often verbally and physically resist the caregiver's efforts to put on the mask. Consequently, they develop conditioned anxiety because of poorly fitting equipment and repeated association of the sight, sound, and sensation of CPAP with discomfort from the mask, physiologic arousal from struggling, or both.³⁵

When treatment failure occurs in the course of OSAS treatment with CPAP, adherence is the major cause, accounting for up to 92% in one study.³² Earlier studies on the efficacy of CPAP in childhood OSAS used subjective self-report and parent-report measures of CPAP usage, and reported a high rate of adherence.^{32,36} More recently, usage recordings by the CPAP machine, which can be digitally downloaded, have enabled objective assessments of adherence and have demonstrated CPAP adherence to be frequently suboptimal. Table 1 summarizes studies investigating CPAP adherence in children.

In a randomized double-blind trial comparing the efficacy and adherence of CPAP and BPAP in PAP-naïve children with OSAS, intention-to-treat analysis that included 8 of 29 patients who failed to return for CPAP usage downloads found adherence for all participants to be suboptimal, at an average usage of 3.8 ± 3.3 hours per night.²⁷ Another study by O'Donnell and colleagues³⁷ reported similar findings of suboptimal adherence, with mean usage of 4.7 hours per night. These results are similar to the findings of a group of Australian investigators who also found a mean CPAP use of 4.7 hours in their study evaluating patterns of CPAP adherence during the first 3 months of treatment in children.³⁸ However, it should be noted that although these numbers are close to or higher than the adult criteria for satisfactory CPAP adherence³⁹ (ie, at least 4 hours/night on 70% of nights), children have a longer sleep duration than adults, and thus thresholds for "adequate" levels of adherence may actually be higher. Furthermore, a study by Marcus and colleagues⁴⁰ suggested that longer duration of CPAP use (mean minutes used per night) correlates inversely with Epworth Sleepiness Scale scores.

It should also be noted that data collected in the context and controlled setting of a study may not reflect typical levels of adherence in practice settings, in which monitoring, feedback, and support levels are likely to be less intense. This aspect represents a particular challenge for clinicians caring for children with OSAS in clinical practice. Even in an experimental setting, one study reported a dropout rate of 24% (7 of 29) during the course of an investigation that provided free equipment and comprehensive social and technical support.⁴¹ It can be theorized that the reluctance to continue with CPAP treatment in the clinical setting, where resources may be limited, is even more prevalent.

FACTORS INFLUENCING CPAP ADHERENCE

Literature from studies on adult CPAP adherence identified categories of factors that influence or predict adherence to CPAP use: (1) disease characteristics; (2) patient characteristics; (3) treatment titration procedures; (4) technological device factors and side

effects; and (5) psychological and social factors. A summary of these factors and their relationship to the course of CPAP treatment is summarized in Table 2.⁴²

Although these categories provide a framework to better understand patients' decisions to adhere to CPAP treatment, one cannot simply extrapolate and assume that these factors are important in the pediatric population. Several retrospective and observational studies also implicate some of these factors in poor CPAP adherence in the pediatric population. However, although only a few prospective studies,^{43,44} discussed herein, are available, these seem to demonstrate a different profile of factors from those shown to predict poor adherence in adults.

Psychosocial Factors

One prospective study collected data potentially pertaining to CPAP adherence and correlated these with objective CPAP adherence data. In their cohort of 56 children aged 2 to 16 years, DiFeo and colleagues⁴³ evaluated patient characteristics (obesity, race, gender), disease characteristics (severity of OSAS as evidence by AHI), child characteristics (presence/absence of developmental delay and attention-deficit/hyperactivity disorder), CPAP-related factors (CPAP pressure and nasal symptoms), and psychosocial factors (maternal education, social support). Financial/insurance barriers were controlled for by providing all of the participants with free equipment and treatment.⁴³ Their analysis revealed that the greatest predictor of CPAP use was maternal education ($r = 0.290$, $P = .033$ for mean hours used per night). Disease-specific factors such as polysomnographic measures and symptoms of excessive daytime sleepiness, which predict CPAP adherence in the adult population, did not significantly correlate with poor CPAP use, nor was there predictive value found in clinical factors such as CPAP pressure and nasal symptoms. The investigators postulated that higher maternal education provided better parental understanding of the consequences of OSAS and the importance of treatment adherence.

DiFeo and colleagues⁴³ also found the Medical Outcomes Study Social Support (MOSS) questionnaire, a measure of family social support, to be somewhat predictive of CPAP adherence, though not as strongly as maternal education. This finding suggests that a combination of caregiver (in this case mothers) knowledge and social support are both important to adherence in children. Several empirical investigations have found that patients' level of adherence is positively associated with their motivation to achieve or maintain good health. Cobb and Jones⁴⁵ suggested that social support encompasses: (1) the supportive behavior of individuals family and friends; (2) the nature of the social network surrounding individuals; and (3) the individual's perceptions of the support provided by their family and friends. However, social support may also exert a stressful influence on the patient when the level of support is perceived as threatening to the patient's autonomy.⁴⁶ This influence is likely to be of greatest importance with adolescent patients.

Patients' Characteristics

In the aforementioned prospective study that evaluated risk factors for pediatric CPAP adherence, DiFeo and colleagues⁴³ also found that certain patient characteristics predicted poor CPAP adherence. African American children, independent of socioeconomic status,

were less likely to be adherent to CPAP than children of other races. This finding mirrors observations found in adult CPAP adherence literature.⁴⁷ Older children, particularly adolescents, were also less likely to adhere to CPAP therapy, which is consistent with data from studies of adherence to other pediatric medical regimens (see later discussion).^{48–50}

Device Factors

Most studies have shown that the type of CPAP device used does not affect adherence, suggesting that adherence is affected by general factors more than by device-specific or disease-specific factors. However, one pediatric study showed that full-face masks were associated with lower adherence than nasal masks.³⁷ Studies in both children and adults comparing different types of pressure delivery, such as BPAP or C-Flex/BiFlex (which deliver a slight decrease in pressure during early expiration and/or inspiration), have had mixed results, but have generally not shown an improvement in adherence in comparison with standard CPAP.^{41,51,52}

Factors Affecting Adherence in Adolescents

Studies on adherence to other medical regimens for the treatment of asthma, epilepsy, and diabetes have shown poor adherence in adolescents.^{53,54} In their review of factors influencing adherence to medical treatments, Fotheringham and Sawyer⁴⁶ observed lower adherence in adolescents than in adults. These investigators argue that poor adherence may be due to the struggle for greater autonomy at home, and reflects rebellion against the regimens' control over patients' lives.

There is a paucity of data describing CPAP adherence in adolescents. Prashad and colleagues⁵⁵ investigated CPAP adherence in 21 adolescents using qualitative semistructured interviews that explored issues of stress, parenting style, family structure and organization, behavioral and emotional problems, knowledge of OSAS, and benefits of CPAP, combined with usage data from the CPAP machine. Their qualitative results found that patients/families with high CPAP usage had stable family structure, high knowledge of CPAP, free communication between parent and adolescent, motivation from a desire to please the caregiver, and an "authoritative parenting style"⁵⁶ whereby children are encouraged to inquire about rules to fully understand them. In this context, parents explained the importance of CPAP use and gave adolescents the choice to use CPAP or not. Adherence was also higher in patients who were prescribed CPAP before adolescence. This finding was postulated to be related to the longer opportunity for CPAP to become a part of the patient's routine when it is prescribed at a younger age, instead of in adolescence when issues of rebellion and emerging autonomy may prove challenging. It was concluded that health education and family involvement are important components in promoting CPAP adherence, and that support strategies should be tailored to the individual and take into account the developmental process of adolescence.

OVERCOMING BARRIERS TO ADHERENCE

Child/Parent Engagement

Successful promotion of CPAP adherence infers addressing child/parent engagement in the initial acceptance of CPAP. Some barriers to this include preconceived notions about CPAP, and outcome expectations incongruent to that of the treating physician and staff.⁵⁷

Determination of the child's and parent's preexisting attitudes toward CPAP is an important first step, and functions to dispel disbeliefs and tailor patient education on outcome expectations. Central to this process of engaging the child and the parent is maintenance of a positive and supportive approach by everyone involved. Such an approach is especially important in children with complex medical problems, for whom gradual CPAP exposure in a supportive setting and anticipatory guidance for troubleshooting may be particularly important.⁴²

Identification of Specific Barriers

To improve adherence, it is vital to identify specific barriers to adherence that both families and patients themselves experience. A significant portion of the literature on adherence has focused predominantly on the concerns of the patients' family alone.⁵⁸ To address this gap, Simon and colleagues⁴⁴ constructed a psychometric questionnaire to evaluate child and family barriers to CPAP adherence. The Adherence Barriers to CPAP Questionnaire (ABCQ) included 31 questions developed across several conceptually derived domains including side effects, time management, health care provider relationships, equipment concerns, and family support, to name a few. The ABCQ demonstrated excellent reliability and validity, and correlated with objective adherence rates. Having a greater amount of barriers identified on ABCQ was associated with poorer rates of adherence for both parent ($r = -0.44$, $P = .002$) and the child ($r = -0.44$, $P = .002$). Table 3 summarizes the most frequently endorsed barriers to CPAP reported. The ABCQ represents the first tool specifically to assess specific barriers to CPAP, but further investigation is needed to determine its utility as a measure of treatment outcomes.

Initial Acceptance

An unpleasant initial exposure to CPAP has been observed to lead to a prolonged period of treatment rejection from both child and caregiver.⁵⁷ Patients with tactile aversions (eg, children with developmental delays) are particularly challenging because they are not typically accepting of the mask at the time of initial presentation. Families and medical practitioners must be prepared to invest substantial time and patience toward successfully initiating and maintaining CPAP therapy.

Overcoming the child's aversion to CPAP may require desensitization procedures that allow the child to slowly habituate by gradual exposure to the mask and machine (ie, operant conditioning). An example would be to divide the procedures into daytime and nighttime practices in an attempt to establish CPAP as part of the child's routine activities.⁵⁹ This approach is further detailed in Table 4. Each practice session should start with an enjoyable, calming activity so the child begins to associate CPAP with positive experiences. The child should be offered small rewards such as stickers or a prize each time he or she is successful

with the practice session, in addition to a verbal praise when the child complies with the parental request. Any negative behaviors such as crying or yelling are ignored. For older children or adolescents, a token economy can be implemented, designed to increase desirable behavior and decrease undesirable behavior with the use of tokens, such as stickers or check marks. The child should receive a sticker immediately after displaying desirable behavior (eg, complying with a request to put the CPAP mask on for designated amount of time). The stickers are collected (or counted up on a chart) and later exchanged for a meaningful object or privilege. Emphasis is placed on practice, patience, and persistence for the overall implementation of CPAP. The goal is to avoid struggles and negative interactions around CPAP, and to implement CPAP in the least stressful way possible for children and parents.

The location where CPAP initiation occurs has also been observed to have an effect on adherence to therapy. Ramirez and colleagues⁶⁰ evaluated long-term adherence to CPAP among 62 children with OSAS who were followed in a pediatric CPAP/noninvasive unit. A high level of adherence was observed in patients who initiated CPAP and followed up in the unit. The investigators postulated that the high level of adherence may have been due to the availability of a dedicated team that provided positive and continuous support for the patient and family from CPAP initiation through follow-up. However, most patients can successfully undergo CPAP initiation in the outpatient setting.

Behavioral Intervention

Behavioral conditioning may be a useful tool to help achieve CPAP acceptance. Koontz and colleagues³⁵ studied 20 children with OSAS, aged 1 to 17 years, who were referred from their respective primary physicians for CPAP nonadherence. Patients and their guardians were then allowed to self-select into 1 of 3 treatment arms: (1) behavior therapy; (2) behavior consultation and recommendation uptake; (3) behavior consultation without recommendation uptake. Before assignments into their respective treatment arms, all children and their guardians attended a behavior consultation with trained staff. This consultation sought to identify individual information about the child's preferences and dislikes to generate recommendations. Results of the study showed that patients in both behavior therapy (75%) and behavior consultation with recommendation uptake (100%) had a statistically significantly heavier CPAP use than those in the behavior consultation group without recommendation uptake (0%). A graphic representation of the results is shown in Fig. 2. This retrospective and descriptive interventional study suggests that both brief behavior consultations and extensive behavioral therapy may offer effective and relatively inexpensive interventions for CPAP adherence.

Side Effects

Similar to any form of treatment, CPAP has associated side effects that may have a negative impact on adherence if unaddressed. Nasal symptoms are the most common side effect, caused by the drying effects of CPAP by altering nasal mucosa and impeding mucociliary clearance.^{30,61} This symptom can be ameliorated with a heated humidifier, maintenance of clean equipment, and, in some cases, intranasal steroids, saline, antihistamines, montelukast, or other treatments.^{31,62} Skin irritation and even ulceration can occur from a tight-fitting

mask or accumulation of skin oils and debris from poor mask maintenance; rarely, allergies to the mask may occur.⁵⁷ A properly fitted mask and daily cleaning of the equipment is thus integral to CPAP adherence. Central apnea at high pressures can be a complication of CPAP use in some children because of an active Hering-Breuer reflex.⁶³ Switching from CPAP to BPAP with a backup rate addresses this problem.

AN EXAMPLE OF A CLINICAL APPROACH

The Sleep Center at The Children's Hospital of Philadelphia (CHOP) is a tertiary referral center for children with a broad range of sleep disorders (Fig. 3, see Table 4). The center comprises a multi-disciplinary team of sleep physicians, behavioral psychologists, respiratory therapists, and nurses, collaborating to provide comprehensive evaluation and management of patients with various sleep disorders, including OSAS. A general description of the CHOP PAP protocol as a model of care follows.

The sleep physician's role in the initial CPAP visit is to educate the family about OSAS, its pathophysiology, complications, and the importance of treating it. The basic mechanism of how CPAP works and side effects that might be encountered are also discussed. The respiratory therapist and nurses present different types of mask interfaces from which the family/patient can choose. The patient is encouraged to try on several masks to determine which is the most comfortable (Fig. 4). Having the patient and family involved in choosing the mask offers them an opportunity to increase a sense of control in a situation where they may feel a loss of control. The psychologist assesses the patient's and caregiver's attitudes and beliefs about CPAP, potential barriers to using CPAP, in addition to family functioning and patient and family routines. He or she then discusses behavioral techniques (eg, mask desensitization, operant conditioning) to help improve adherence, as well as strategies to implement CPAP into the specific patient's daily routine. For example, for a family whose household is chaotic or whose child does not have a "typical routine," the psychologist and family may together develop a specific evening and bedtime routine that ends with putting on the CPAP.

For an adolescent, it may be beneficial to discuss a division of responsibilities between the adolescent and caregiver to reduce feelings in both parties of being overburdened. A caregiver may agree to remind the adolescent to get his or her CPAP ready before bed and to help with the deeper weekly cleaning of the equipment, while the adolescent agrees to be responsible for putting the CPAP on each night and participate in the daily care of the equipment (eg, pouring water out of the humidifier, rinsing the humidifier, tubing, and mask each day, and setting out to air dry in the morning). This strategy aims to respect and improve the autonomy and responsibility of the adolescent while providing the necessary support he or she likely needs in adhering to a medical intervention without a sense of being nagged.

A durable medical equipment representative also meets with the family to discuss machine operation, maintenance, and troubleshooting, and to provide equipment for the family to take home that day. At the end of the visit, it is verified that the family is given a network of support by providing contact information for them to call in case of further inquiry. This

process is spearheaded by the CPAP coordinator (a position split between a respiratory therapist and psychologist) to provide an organized channel for subsequent clinic follow-up. A CPAP titration polysomnogram is typically not performed until the child is tolerating CPAP set at a low pressure at home.

Within a week, a phone call is made by a CPAP coordinator to check adherence and to troubleshoot any problems such as device issues or CPAP side effects, as well as to assess behavioral implementation concerns or barriers. If behavioral concerns exist or persist, the psychologist follows up by phone regularly. The respiratory therapist and nurses also provide phone support for technical or side effects and medical concerns as needed. Patients are encouraged to follow up at the clinic after 1 month, at which time adherence is assessed. The CPAP machine's usage data is downloaded to provide an objective measure of the patient's adherence. The results are then discussed with the patient and family, who are shown a visual representation of the usage (Fig. 5). Those who have good adherence are given an incentive CPAP trophy (when developmentally appropriate) (Fig. 6) and have their picture posted on the "CPAP Hall of Fame." Patients with poor adherence are reassessed by the team for specific barriers to adherence such as troubleshooting machine problems, issues with the mask fit or putting it on correctly, or behavioral barriers. Adolescents may need help troubleshooting simple issues, such as finding an electrical outlet near their bed, developing a plan to take their machine with them when staying overnight at relatives' or friends' homes, or setting an alert on their phone to remind them to put on their mask at night. For all patients, technical support and behavioral reinforcement is provided, with close follow-up thereafter. Children are seen on a regular basis, with the frequency of their clinic visits depending on their age, severity of medical condition, and CPAP adherence.

In almost all cases, CPAP is implemented in an outpatient setting. Occasionally a family (typically with an infant, toddler, or developmentally delayed child) is unable to implement a desensitization program, and a short 2- to 4-week intervention with home nursing is implemented. This approach often breaks the cycle of CPAP refusal, anxiety, and frustration, and demonstrates to the family that the child can tolerate CPAP. Typically parents are able to continue implementing CPAP at the end of the nursing intervention. Rarely, patients are admitted to an inpatient unit when all other measures fail.

SUMMARY

CPAP is a safe and effective treatment for children with OSAS that has many beneficial effects. The major limiting factor for successful treatment with CPAP is adherence. Psychosocial factors and patient characteristics are the main predictors of poor adherence, with low maternal education being the strongest. Managing suboptimal adherence entails engaging and educating the child and parent about CPAP. This approach involves discussing preexisting attitudes, outcome expectations, and side effects, in addition to providing constant support from a dedicated team. More intensive cognitive-behavioral interventions may prove useful in overcoming poor CPAP adherence in complex cases.

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KEY POINTS

- Obstructive sleep apnea syndrome (OSAS) is a relatively common disorder affecting approximately 1% to 5% of children aged 5 to 13 years, and is associated with adverse neurobehavioral, cognitive, and physiologic consequences.
- Adenotonsillectomy is considered to be the first-line treatment; however, not all patients are surgical candidates and some patients will continue to have symptoms after surgery.
- Continuous positive airway pressure (CPAP) is a safe and effective treatment for children with OSAS who do not improve with surgery or who are not candidates for surgery. The major limiting factor for successful treatment with CPAP is adherence.
- Psychosocial factors and patient characteristics are the main predictors of poor adherence, with low maternal education being the strongest.
- Managing suboptimal adherence entails engaging and educating the child and parent about CPAP; this involves discussing preexisting attitudes, outcome expectations, and side effects, in addition to providing constant support from a dedicated team.
- More intensive cognitive behavioral interventions may prove useful in overcoming poor CPAP adherence in complex cases.

Fig. 1.

(A) A full-face CPAP mask with headgear attached to a forehead spacer. (B) A CPAP air compressor machine with humidifier. (C) A nasal CPAP mask. (D) A nasal CPAP device with nasal pillows.

Fig. 2.

Results of CPAP adherence strategies in a group of children referred for behavioral counseling for poor CPAP adherence. Subjects were divided into 3 groups: BT received consultation and recommendations plus a course of behavior therapy; CR+ received a 1.5-hour consultation and recommendation session; CR- underwent a consultation and behavior therapy was recommended, but the family did not follow up. The y-axis shows the average number of hours per night of CPAP use. The hours of use are further subdivided into baseline, during treatment, and posttreatment values. Seventy-five percent of children who received behavior intervention (CR+ and BT groups) tolerated CPAP successfully ($P < .5$ for CR+ vs CR- group; other comparisons were not significant). (Adapted from Koontz KL, Slifer KJ, Cataldo MD, et al. Improving pediatric compliance with positive airway pressure therapy: the impact of behavioral intervention. *Sleep* 2003;26(8):1012; with permission.)

Fig. 3.

Example of a CPAP adherence algorithm for patients with obstructive sleep apnea.

Fig. 4.
A patient with trisomy 21 trying on a nasal mask during a CPAP initiation visit.

Fig. 5. CPAP data downloads. The column on the left shows usage data from a nonadherent patient, and the column on the right is from an adherent patient. The x-axis shows 24-hour clock time and the left y-axis shows the date. CPAP use for 4 hours or more is shown in green, less than 4 hours in red; air leak is shown in black. Corresponding numbers are shown on the right y-axis. Air leak may be due to the mask being damaged, fitting poorly, or being off the face when the air compressor is turned on.

Fig. 6.
A patient receiving a CPAP champion trophy for good adherence.

Table 1

Summary of select studies investigating CPAP adherence in children

Authors, Ref, Year	Population	Age	OSAS Characteristics	Associated Conditions	Measure of Adherence	Findings
DiFeo et al, ⁴³ 2012	N = 56 children and their parents 68% male 59% African American 36% Caucasian	2–16 y	AHI 19 ± 16/h Naïve to CPAP	71% obese 23% with neurodevelopmental disabilities 20% with genetic syndromes	Usage data from machine (at 1 and 3 mo)	Average use 3 ± 3 h per night after first month, 2.8 ± 2.7 h on third month Greatest predictor of use was maternal education Older, typically developing African American youth with low social support had poor adherence
Simon et al, ⁴⁴ 2012	N = 51 children and their parents 51% male 51% non-Hispanic Caucasian 37% African American 64% had Medicaid	8–17 y	Average AHI 17/h Average CPAP use of 22.9 mo	73.5% overweight/ obese	Usage data from machine	Poor adherence with average use of 3.35 h per night Questionnaire developed was able to identify specific barriers to CPAP
Marcus et al, ⁴¹ 2006	N = 29 children 72% male 51% African American	2–16 y	Newly diagnosed OSAS	65% obese 10% craniofacial abnormalities 34% systolic hypertension	Parental report and usage data from machine for 6 mo	Average use 3.8 ± 3.3 h per night 9 dropouts Parental report overestimated actual use No difference in adherence between CPAP and BPAP
O'Donnell et al, ³⁷ 2006	N = 50 children	Mean 10 ± 5.1 y 66% male	Median AHI = 11.3	78% with comorbidity	Usage data from machine	Average use 6.3 h per night
Koontz et al, ³⁵ 2003	N = 20 children 55% African American 30% Caucasian	1–17 y	Nonadherent children referred by physicians	45% with some degree of developmental delay	Usage data from machine	3 groups: 1 Consultation (usage 8.58 h/night) 2 Consultation with behavior therapy (usage 5.88 h/night) 3 No consultation/behavior therapy (usage 0.67 h/night)
Massa et al, ³⁶ 2002	N = 66 children 59% male	Infant to 19 y	All moderate to severe OSAS (AHI >5 per hour)	35% craniosynostosis syndromes 9.1% isolated facial defects 6.1% obese 3% trisomy 21 3% cerebral palsy	Parental report	67.7% report good adherence (uses every night and all night long) CPAP tolerated by 86%
Marcus et al, ¹⁷ 1994	N = 94 children 64% male	2 wk to 19 y	—	27% obese 25% craniofacial abnormalities 13% trisomy 21	Parental report	12.7% with inadequate adherence

Abbreviations: AHI, apnea-hypopnea index; BPAP, bilevel positive airway pressure; CPAP, continuous positive airway pressure; OSAS, obstructive sleep apnea syndrome.

Data from Refs. 17,35–37,41,43,44

Table 2

Factors that influence on CPAP adherence in adults

Factor	Relationship to Course of Treatment		
	Pre-CPAP Exposure	Initial CPAP Exposure	Home CPAP Treatment
Disease and patient characteristics	Disease severity Sleepiness Upper airway patency Race Socioeconomic status	Depression Mood Personality type	Depression Mood Personality type
Treatment titration procedure	—	Autotitrating CPAP	—
Technological device factors and side effects	Claustrophobia	Heated humidification	Heated humidification
Psychological and social factors	Disease- and treatment-specific knowledge	Disease- and treatment-specific knowledge	Disease- and treatment- specific knowledge Decisional balance Active coping style Disease-specific risk perception

Abbreviation: CPAP, continuous positive airway pressure.

Adapted from Sawyer AM, Gooneratne NS, Marcus CL, et al. A systematic review of CPAP adherence across age groups: clinical and empiric insights for developing CPAP adherence interventions. *Sleep Med Rev* 2011;15(6):345; with permission.

Table 3

Most frequently endorsed barriers to CPAP reported on the ABCQ

Parent-Report Item (% Agree/Strongly Agree)		Child-Report Item (% Agree/Strongly Agree)	
Does not use when away from home	45.1	Does not use when away from home	47.0
Child not feeling well	44.0	Just want to forget about OSAS	43.1
Forgets	39.2	Not feeling well	42.0
Child does not feel like using CPAP	30.0	Forgets	39.2
Child just wants to forget about OSAS	23.6	No one helps to use CPAP at night	31.4
Child embarrassed about using CPAP	22.0	Embarrassed about using CPAP	29.4

Abbreviations: ABCQ, adherence barriers to CPAP questionnaire; CPAP, continuous positive airway pressure; OSAS, obstructive sleep apnea syndrome.

Adapted from Simon SL, Duncan CL, Janicke DM, et al. Barriers to treatment of paediatric obstructive sleep apnoea: development of the adherence barriers to continuous positive airway pressure (CPAP) questionnaire. *Sleep Med* 2012;13(2):172–7; with permission.

Table 4

Desensitization procedure for infants and school-aged children

Daytime Practice	Nighttime Practice
<p>Introducing the mask</p> <p>The child and parent play together with the mask and machine. The parent puts the PAP on a stuffed animal, doll, themselves, etc</p> <p>Place the mask (not attached to the hose/ machine) on your child's face for 5 s</p> <p>Place the mask (not attached to the hose/ machine) on your child's face for 10 s</p> <p>Place the mask (not attached to the hose/ machine) on your child's face for 1 min</p> <p>Place the mask on your child's face and connect the headgear on <i>both</i> sides for 1 min</p> <p>Repeat above step, slowly increasing the amount of time your child wears the mask and headgear each practice session</p>	<p>Adjusting to the sounds at night</p> <p>While your child is going through desensitization practice during the day, begin turning on the PAP machine at night without attaching it to your child so he/she can get used to the noise</p>
<p>Turning on the air</p> <p>Place the mask and headgear on your child; attach the hose to the mask and to the machine, and turn the air on for 5 s</p> <p>Repeat above step, slowly increasing the amount of time each practice session</p> <p>Wearing the mask with airflow while lying down</p> <p>Have your child lie down on the bed or couch.</p> <p>Place the mask (attached to the hose and both sides of the cap) on your child's face for 5 min</p> <p>Have your child lie down on the bed or couch.</p> <p>Place the mask (attached to the hose and both sides of the cap) on your child's face for 10 min</p> <p>Have your child lie down on the bed or couch.</p> <p>Place the mask (attached to the hose and both sides of the cap) for your child's face or 15 min</p>	<p>Making PAP part of your child's bedtime routine</p> <p>Add PAP to your child's bedtime routine.</p> <p>Have your child try to fall asleep after you place the mask (attached to the hose on both sides of the cap) on the face and the air is turned on for 15 min</p> <p>Continue nightly until your child is able to fall asleep with PAP. If your child cannot fall asleep with PAP, do 15 min of practice</p> <p>Continue daytime practice and bedtime practice until your child is able to fall asleep with PAP on and sleep with it for most of the night. Replace PAP at night if it falls off or is removed</p>

Abbreviation: PAP, positive airway pressure.

Adapted from The Children's Hospital of Philadelphia. CPAP education: noninvasive ventilation for infants and children. 2013. Available at: <http://www.chop.edu/service/sleep-center/cpap-education/non-invasive-ventilation-for-infants-and-children.html>. Accessed November 25, 2013; with permission.