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Original Article

Evaluation of the efficacy of a dedicated table to improve CPAP adherence in children: a pilot study

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ABSTRACT

Objective: Only a few studies have addressed strategies to improve continuous positive airway pressure (CPAP) adherence in children with obstructive sleep apnea. The aim of our study was to assess the efficacy of a table based on token economy to improve CPAP adherence.
Methods: A table was proposed to children nonadherent to CPAP (<3 h per night of CPAP use), eight days after CPAP initiation (D8). The child has to fill the table on a daily basis with green (I used my CPAP this night) or red (I did not use my CPAP) tokens. Objectives of CPAP use and rewards were decided between the child and their parents. An assessment of CPAP adherence was performed one month after initiation of table filling (M1) and compared to CPAP adherence at D8, and to data of adherent children.
Results: Data of six nonadherent and nine adherent children were gathered (age 5 ± 5 vs. 5 ± 3 years, $p = 0.953$; apnea–hypopnea index 20 ± 15 vs. 25 ± 16 events/h, $p = 0.550$, respectively). Mean CPAP adherence at D8 was 4.7 ± 1.6 nights/wk and 1h00 ± 0h33 in the nonadherent children, vs. 6.9 ± 0.4 nights/wk and 7h16 ± 1h51 in the adherent group ($p < 0.01$). The mean number of nights per week at M1 was 6.4 ± 0.6 nights in nonadherent children ($p = 0.086$), and was equivalent to that in adherent children (6.8 ± 0.6 nights, $p = 0.126$). Mean adherence at M1 increased to 4h31 ± 1h12 in non-adherent children ($p < 0.001$), but was still lower when compared to that in adherent children (7h27 ± 2h00, $p = 0.007$).
Conclusion: The study findings imply that use of a simple table by a child appears to be effective in improving CPAP adherence at one month.

1. Introduction

In children with obstructive sleep apnea syndrome (OSAS), continuous positive airway pressure (CPAP) has proved its benefit by reducing the apnea–hypopnea index (AHI) and by improving neurobehavioral outcomes, such as attention deficits, daytime sleepiness, behavior, and caregiver- and child-reported quality of life, even in case of suboptimal adherence [1]. However, Marcus et al. [1] found a significant correlation between the decrease in sleepiness at three months assessed using the Epworth Sleepiness Scale and CPAP adherence. This study also highlighted the fact that clinicians should encourage any CPAP use, as reinforcing CPAP use is beneficial in many domains of the child's daily life.

Several studies have investigated the objective adherence to CPAP in children with OSAS and reasons for adherence difficulties [2–10]. DiFeo et al. [5] found, as in other studies of adherence to pediatric medical regimens, that maternal education was strongly associated with adherence to CPAP therapy. They presumed that this was due to a better parental understanding of the consequences of OSAS and the importance of treatment. They also reported that older children and adolescents were less likely to be adherent to CPAP than younger children [3,5]. However, children with developmental disabilities did not show age-related differences in adherence, probably because parents were more closely involved in their care. Recently, Xanthopoulos et al. [10] examined the relationship between caregiver and patient-reported health cognitions about CPAP. They found that caregiver CPAP-specific self-efficacy was an important factor to consider when starting

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youth on CPAP therapy for OSAS. They concluded that using strategies to improve caregiver self-efficacy, beginning at CPAP initiation, may promote CPAP adherence.

Only a few studies have addressed strategies to optimize adherence to CPAP in children [11–17]. These strategies are based mainly on educational, technological, psychosocial (behavioral), pharmacological, and multi-dimensional approaches [16]. Some of the interventions have been effective in improving CPAP use, whereas other failed; however, as suggested by Sawyer et al. [16], the clinical applicability and cost-effectiveness of any intervention must also be carefully examined.

In an attempt to propose a simplified tool that would be easily implementable in any center and with low costs, we proposed a dedicated table to improve CPAP adherence, based on the token economy system, in children naive to CPAP and nonadherent to their treatment. This table aimed to reinforce the involvement of both the parents and children, as objectives of CPAP use and rewards were planned together. The aim of our study was to assess the efficacy of the table on CPAP adherence in nonadherent children after one month, and to compare the findings with adherent children followed within the same time period.

2. Methods

2.1. Patients

Data of all consecutive children with OSAS followed at Gonesse Hospital, in France, between January 2015 and March 2016, who were nonadherent to their home CPAP therapy and to whom supportive intervention was proposed, were retrospectively analyzed. Data of children followed within the same time period and with a CPAP adherence over 4 h per night were gathered. All the children had a polysomnography prior to CPAP initiation. CPAP was initiated in case of an AHI >10 events/h or >5 events/h associated with clinical symptoms. CPAP was proposed prior to scheduled ear–nose–throat (ENT) surgery, in case of persistent OSAS despite previous ENT surgery or in children in whom ENT surgery was not indicated. CPAP was initiated during inpatient hospitalizations.

Data of the adherent children were selected to determine a similar mean age and mean AHI between the two groups. All the children were followed at home for CPAP therapy by the same home care provider. Clinical data and AHI of the two groups were gathered.

The study was approved by the Institutional Review Board of the French Learned Society for Respiratory Medicine, Société de Pneumologie de Langue Française (CEPRO 2017-033), and all the parents gave informed consent for the CPAP evaluation of their child.

2.2. Procedure

The follow-up procedure consisted of two home visits within the first month of CPAP therapy, by the home care provider. The first visit was performed eight days after CPAP initiation (D8), whereas the second visit was performed about one month after. Adherent and nonadherent children were followed as per routine practice, with a careful examination of all the equipment (device, interface, circuit, and humidifier), a further explanation of the use of the device to the parents and/or child if necessary, and the download of the in-built software data.

In children in whom adherence to CPAP was <3 h per night at D8, a dedicated table was proposed with the aim to follow and improve CPAP use. This table was designed by a staff member of the home care provider company (A.M.-R.), and is a monthly calendar table based on the token economy system. It consisted on giving the children two kinds of tokens: a green token to be fixed daily on the table to indicate that the CPAP device was used during the night, and a red token to indicate that the CPAP device was not used. Alternately, the child could also use green and red pencils to draw a circle on the corresponding cell of the table. The child was asked to daily fill the table according to his/her CPAP use. No objective of night duration of CPAP use was fixed; however, an objective of number of nights per week with CPAP was determined together with the child and his/her parents, and a reward was chosen. The reward could be a visit at the zoo, a toy, etc. The child was given the reward once the home care provider checked the table at M1 and attested to the progress.

Adherence to CPAP (number of hours of CPAP use, number of nights per week with CPAP, and percentage of day use within the selected period) was assessed using the in-built software data of the CPAP device. Adherence was determined one month after the table was initiated (M1), and was compared to adherence at D8. Adherence was also compared between nonadherent and adherent children.

2.3. Statistical analysis

Data were presented as mean ± standard deviation. Comparisons between the two groups were done using the unpaired *t* test in case of a normal distribution, or the Mann–Whitney rank sum test otherwise. Comparisons within the same group were done using the paired *t* test in case of a normal distribution, or the Wilcoxon signed rank test otherwise. A p value of <0.05 was considered as significant.

3. Results

Data of six nonadherent children and nine adherent children were gathered (Table 1). Two children had Down syndrome, with the majority of patients having adenoids and/or tonsillar hypertrophy with no developmental delay. The mean age was 5 ± 5 vs. 5 ± 3 years ($p = 0.953$), and mean baseline AHI was 20 ± 15 events/h vs. 25 ± 16 events/h ($p = 0.550$), respectively. Mean CPAP level was 6.2 ± 1.2 cm H_2O vs. 6.4 ± 1.0 cm H_2O ($p = 0.632$), respectively. Fig. 1 presents an example of filled tables by patients #3 and #5. Table 2 summarizes the objective number of nights per week with CPAP and the rewards determined by the children and their parents.

The mean number of nights per week with CPAP at D8 was 4.7 ± 1.6 nights in the nonadherent children vs. 6.9 ± 0.4 nights in the adherent group ($p = 0.006$). The mean number of hours per night with CPAP at D8 was 1h00 ± 0h33 in the nonadherent children vs. 7h16 ± 1h51 in the adherent group ($p < 0.001$) (Fig. 2). The percentage of day use of CPAP at D8 was 67% ± 23% of day use in the nonadherent children vs. 98% ± 5% in the adherent group ($p = 0.015$) (Fig. 2).

The mean number of nights per week with CPAP at M1 increased to 6.4 ± 0.6 nights in the nonadherent group, even though it was not significantly different when compared to D8 ($p = 0.086$); however, it was significantly similar when compared to the adherent group (6.8 ± 0.6 nights per week, $p = 0.126$). Five of the six nonadherent children reached the predetermined number of nights per week with CPAP (Table 2). Patient #5 was the only child with an objective of seven nights per week, because of daytime sleepiness, but used CPAP only six nights per week. The mean number of hours per night with CPAP improved significantly at M1 (4h31 ± 1h12) in the nonadherent group when compared to D8 ($p < 0.001$) (Fig. 3), but was still significantly lower when compared to that in the adherent group (7h27 ± 2h00, $p = 0.007$). The percentage of day use of CPAP at M1 increased to 92% ± 9% in the non-adherent group, even though it was not significantly different when compared to D8 ($p = 0.086$); however, it was significantly equivalent when compared to the adherent group (97% ± 8%, $p = 0.172$).

Table 1
Characteristics of the study population.

Patients	Sex	Pathology	Age (y)	Height (cm)	Weight (kg)	BMI *z* score	AHI (events/h)	CPAP level (cm H_2O)
Nonadherent								
1	M	Tonsillar hypertrophy	3.7	103	16	−0.61	15	6
2	F	Tonsillar hypertrophy	2.2	98	13	−2.45	7	5
3	M	Tonsillar hypertrophy	2.8	102	17	0.22	16	6
4	F	DS, psychomotor delay	14.1	147	78	2.36	47	8
5	F	Tonsillar hypertrophy	2.9	99	17	1.12	11	5
6	F	Tonsillar hypertrophy	3.0	100	13	−2.94	24	7
Adherent								
1	M	Adenotonsillar hypertrophy	5.0	110	22	1.76	57	6
2	M	Adenoids hypertrophy	6.6	120	28	1.83	10	6
3	F	DS	0.9	76	9	–	33	6
4	F	Tonsillar hypertrophy	3.4	90	11	−2.02	6	5
5	M	Tonsillar hypertrophy	3.0	84	10	−1.82	16	6
6	F	Tonsillar hypertrophy	2.2	90	12	−1.17	29	8
7	M	Tonsillar hypertrophy	2.2	91	15	1.13	39	8
8	M	Tonsillar hypertrophy	8.6	130	27	−0.01	17	7
9	M	Tonsillar hypertrophy	8.6	104	30	2.47	17	6

AHI, apnea–hypopnea index; BMI, body mass index; CPAP, continuous positive airway pressure; DS, Down syndrome; F, female; M, male.

Fig. 1. Examples of tables filled out by two patients. The table had to be filled in on a daily basis (Monday to Sunday) during four weeks (weeks 1–4). (Left side) Patient #5: Objective: I use my continuous positive airway pressure (CPAP) device every night; Reward: a toy. (Right side) Patient #3: Objective: I use my CPAP device six days a week (I choose the day off); Reward: a visit at the zoo and a dinosaur toy. (Green drawing) I used my CPAP device this night. (Red drawing) I did not use my CPAP device this night. (For interpretation of the references to color in this figure legend, the reader is referred to the Web version of this article.)

Table 2
Objective CPAP adherence and rewards determined with nonadherent children and the parents.

Patients	Objective no. of nights per week	Rewards at M1	No. of nights per week at M1
1	6	Go to swimming pool	7
2	6	A toy	7
3	6 (the child chose the night without CPAP)	Visit at zoo and dinosaur toy	6
4	6	Watch a cartoon with her sister	7
5	7[a]	A toy	6
6	6	Dinner at fast food restaurant	7

CPAP, continuous positive airway pressure; M1, one month after table use was initiated.
[a] Because of daytime sleepiness.

4. Discussion

To our knowledge, this study is the first to propose a table based on the token economy system to improve CPAP adherence in children with OSAS. This simple inexpensive table proved its efficacy to enhance adherence to CPAP in nonadherent children after one month, by increasing the number of nights with CPAP per week. However, the number of hours per night with CPAP was still low as compared to that in adherent children.

Components of a successful intervention to promote CPAP adherence in children are likely to address both the child's and his/her parents' engagement in initial acceptance and use of CPAP [16]. In that context, Rains [11], Koontz et al. [12], and Slifer et al. [14] tested a behavioral intervention aimed at improving positive airway pressure (CPAP or bilevel) use in nonadherent children with or without developmental delay and their parents. Their results supported the importance of behavior analysis and therapy for increasing adherence to CPAP in children. Accordingly, our results also supported the idea that, not only the involvement of both the children and their parents is mandatory for the success of therapy, but also that the involvement of the home care provider may be of particular benefit in the success of the treatment. Indeed, a strong collaboration among the patient and family, the health care center, and the home care provider is very important to optimize adherence to therapy [18].

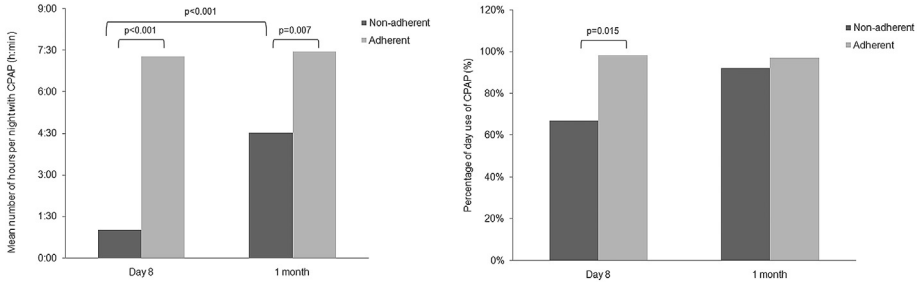

Fig. 2. Continuous positive airway pressure (CPAP) adherence at day eight and after one month in nonadherent and adherent children. (Left side) The mean number of hours per night with CPAP at day eight was low in the nonadherent children and improved significantly at one month. Adherence was lower when compared to that in the adherent group. (Right side) The percentage of day use of CPAP at day eight was low in nonadherent children. At one month, it was significantly equivalent when compared to the adherent group.

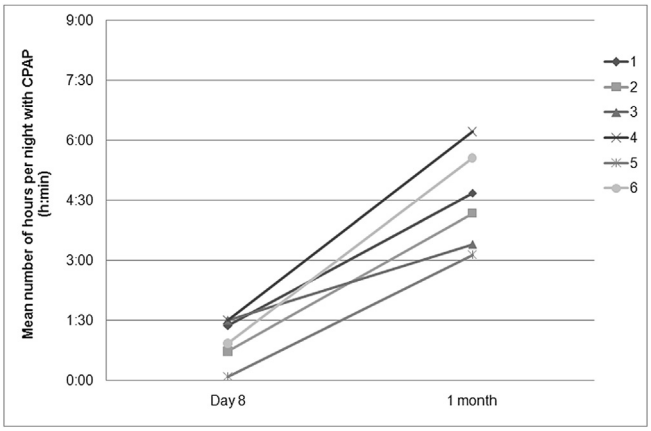

Fig. 3. Mean number of hours per night with continuous positive airway pressure (CPAP) at day eight and after one month with the table in nonadherent children. The mean number of hours per night with CPAP improved significantly at one month when compared to day eight.

The strategy that we used was based on maximizing the number of days of CPAP use per week, rather than the number of hours per night of CPAP use, as we supposed that if using CPAP every night became a sleeping habit for the child, the number of hours of use could increase with time. Indeed, CPAP has proved its benefit even in cases of suboptimal adherence [1], highlighting the fact that daily use may be very important despite a low number of hours per night according to the child's sleep time. As a fact, the number of hours with CPAP use per night increased using the table in our nonadherent children, even though it was still lower than in adherent children. It is possible that with long term follow-up, the number of hours per night would have increased further. However we could not assess CPAP adherence on a long-term basis, as a majority of our patients were scheduled for ENT surgery and therefore CPAP was discontinued.

We proposed the table only eight days after CPAP initiation, as it appears that monitoring adherence in the first week of treatment and intervening in cases of low adherence may improve long-term CPAP use [4]. However, the table can also be proposed to enhance adherence, in preschool and school-aged children, preadolescents, and children with developmental delay of any age, such as those with Down syndrome, immediately when CPAP is initiated. We do not believe that this table is adapted to adolescents without developmental delay; although, this is unknown and may deserve further study. In our daily practice, the table is now used immediately at CPAP initiation with success on D8 and M1 adherence (data not shown).

Our study has several limitations. First the number of patients was low, and only two patients presented with developmental delay. Second, our population consisted mainly of very young children, so one could suppose that the adherence was based mainly on the parents' will. However, even at a younger age, such as in our youngest, 2.2-year-old patient, the child was able to understand that putting the "elephant trunk" on her face to sleep each night was very helpful to breathe better during sleep, and that she had to fill out the table every day with a majority of green tokens to get the chosen reward. During the study period, there were no school-aged children or adolescents who were nonadherent, and thus no data regarding the impact of this intervention were collected in this age group. Finally, only short term follow-up was assessed, as a majority of our patients were initiated on CPAP prior to scheduled ENT surgery.

5. Conclusion

In conclusion, in this study, the involvement of the child, even at a very young age, with his/her CPAP treatment was able to improve adherence to the treatment at one month, using a simple table. Further studies are required to assess the effectiveness of this table in children, with or without developmental delay, on long-term adherence to CPAP or noninvasive ventilation (NIV). Moreover, studies should address strategies to optimize CPAP/NIV adherence in adolescents, such as CPAP/NIV initiation during sessions for adolescent groups, as adherence to medical treatment in this population is particularly difficult. There is a great need for further research in this area, as CPAP/NIV becomes more widespread in the care management of children.

Conflicts of interest

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.08.032.

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