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SLEEP BREATHING PHYSIOLOGY AND DISORDERS • SHORT COMMUNICATION

Long-term adherence to ambulatory initiated continuous positive airway pressure in non-syndromic OSA children

M-P. Perriol[1] · I. Jullian-Desayes[2,3] · M. Joyeux-Faure[2,3] · S. Bailly[2,3] · A. Andrieux[4] · M. Ellaffi[1] · F. Jounieaux[5] · J-L. Pépin[2,3] · C. Lamblin[5]

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Abstract

Purpose In children, the usual indications for continuous positive airway pressure (CPAP) are residual OSA after adenotonsillectomy and/or persistent OSA due to obesity. Data concerning adherence (hours/night) following ambulatory CPAP initiation are scarce.

Methods An observational cohort of 78 children was followed over 2 years. All exhibited sleep-disordered breathing (SDB) symptoms, were assessed by polysomnography, and prescribed CPAP. CPAP was initiated at hospital for 10 children.

Results OSA children, mean age 10.4 ± 3.2 years, were mostly males (75.6%), with a mean body mass index of 21.2 ± 7.3 kg/m^2, and mean apnea+hypopnea index of 12.2 ± 10.6 events/hour. Seventy-two children were still on CPAP at 3 months, 63 at 6 months, 55 at 1 year, and 34 at 2 years. CPAP was discontinued thanks to rehabilitation programs, dento-facial orthopedics, and/or weight loss. Mean CPAP adherence at 1, 3, 6, 12, and 24 months was respectively 6.1 ± 2.8, 6.2 ± 2.6, 6.2 ± 2.8, 6.3 ± 2.8, and 7.0 ± 2.7 h/night. There was a trend towards higher CPAP adherence and younger age, primary versus middle/high school attendance, higher baseline apnea+hypopnea index, and neurocognitive disorders.

Conclusion In our population, mean CPAP adherence defined in hours per night was high and did not decrease during the 24-month follow-up. These findings support the feasibility of ambulatory CPAP initiation in non-syndromic OSA. The high CPAP adherence is expected to be associated with improvements in neurocognition, and in metabolic and cardiovascular parameters.

Keywords CPAP adherence · OSA · Children · Ambulatory

Introduction

In children, the prevalence of obstructive sleep apnea (OSA) is estimated between 2 and 10% [1]. In non-syndromic OSA (i.e., without neuromuscular disease or craniofacial malformation), the most frequent symptoms are snoring, restless sleep, witnessed apneas, mouth breathing, neurobehavioral disturbances, and cardiometabolic comorbidities with somatic growth retardation in severe cases. In children, weight loss, treatment of nasal obstruction and upper airway inflammation, adenotonsillectomy, and the rapid introduction of maxillary expansion or orthodontic appliances are employed in a stepwise fashion with the goal of achieving complete resolution of OSA. The most common indications for CPAP are residual OSA after adenotonsillectomy and/or persistent OSA in obese children. As in adults, CPAP generally suppresses nocturnal and daytime symptoms and might reduce metabolic syndrome and nonalcoholic fatty liver disease [2], as well as systolic blood pressure [3] although no randomized controlled trial is available in children. Ideally, CPAP titration is recommended during polysomnography in a sleep clinic [4, 5]. Data regarding the efficacy of home-based CPAP titration for children are lacking. CPAP adherence by children varies considerably

✉ I. Jullian-Desayes
ijulliandesayes@chu-grenoble.fr

1 Unité Veille-Sommeil, Hôpital Jean Bernard, Valenciennes, France

2 Pôle Thorax et Vaisseaux, Explorations fonctionnelles cardiorespiratoires, CHU Grenoble Alpes, Grenoble, France

3 Laboratoire HP2, INSERM U1042, Université Grenoble Alpes, Grenoble, France

4 Pôle d'Exploration des Apnées du Sommeil, Nouvelle Clinique Bel air, Bordeaux, France

5 Hôpital Privé de La Louvière, Lille, France

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between studies [4, 6, 7], and predictors of non-adherence are probably specific to this population. To the best of our knowledge, data concerning long-term CPAP adherence and factors impacting CPAP adherence in children are scarce. Thus, the primary objective of this study was CPAP adherence and its long-term evolution (24 months) in non-syndromic OSA children following the initiation of CPAP treatment at home or in ambulatory care during a daytime therapeutic education session. A secondary objective was to determine the predictive factors of CPAP adherence (or non-adherence).

Material and methods

Patients were prospectively recruited by a multidisciplinary network of centers (Lille, Valenciennes, Bordeaux; France) synchronizing their efforts to improve the care of children suffering from non-syndromic OSA. Written informed consent was requested from both parents. CPAP treatment was initiated strictly during a home visit by a nurse or a technician, after an ambulatory daytime therapeutic education session including the child and parents or overnight in hospital with either automated-mode (autoCPAP) or a fixed pressure device. Clinical visits at the sleep clinic were made at 1, 3, 6, 12, and 24 months and the apnea-hypopnea index (AHI) was reassessed by polysomnography at months 12 and 24 to inform the continuation or stop of CPAP.

Continuous data are presented as mean ± SD and categorical data as frequency and percentage. A mixed effect model with a random patient effect was performed to study CPAP adherence, and AHI. Statistical analyses were performed with SAS 9.4.

Results

The cohort of 78 children aged 10.4 ± 3.2 years (5 to 17 years), with sleep-disordered breathing (SDB) symptoms and diagnosis confirmed by a polysomnography, and indicated for CPAP treatment were followed for 2 years (see flow chart Fig. 1). Apneic children were mainly males (75.6%), with a mean body mass index (BMI) of 21.2 ± 7.3 kg/m^2. Among them, 81.8% were snorers, 72.7% had neurocognitive dysfunction, 84.4% had daytime fatigue, and 36.4% had a family history of OSA. From baseline polysomnography, the mean AHI was 12.2 ± 10.6 events/hour and mean SpO_2 was 96.5 ± 1.4%.

CPAP treatment was initiated at home for 42 children, after daytime therapeutic education for 26 other children or in hospital for 10 children. Data were available for 72 patients up to 3 months, 63 up to 6 months, 55 up to 1 year, and 34 up to 2 years (Fig. 1). A significant number of children exhibited

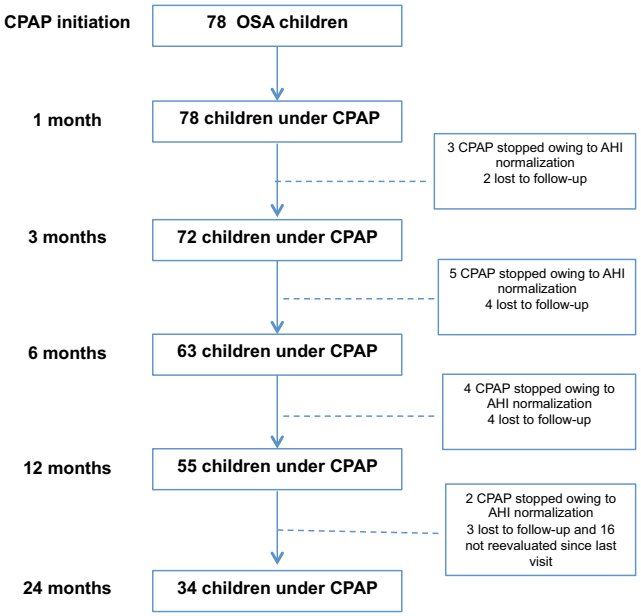

Fig. 1 Study flow chart

resolution of their symptoms and an AHI ≤ 2/h thanks to the efficacy of alternatives to CPAP treatment, i.e., dento-facial orthopedics ($n = 1$) or adenotonsillectomy ($n = 9$). Only 13 (16%) were lost to follow-up (2 at 3 months, 4 at 6 months, 4 at 12 months, and 4 at 24 months). Moreover, the last 20 children to be included have not yet been evaluated at 24 months. Mean CPAP adherence at 1, 3, 6, 12, and 24 months was respectively 6.1 ± 2.8, 6.2 ± 2.6, 6.2 ± 2.8, 6.3 ± 2.8, and 7.0 ± 2.7 h/night (Fig. 2). The mean nightly duration of CPAP use did not decrease during long-term follow-up. In univariate analysis, there was a trend towards an association between high CPAP adherence in hours per night and younger age ($p = 0.08$), high AHI at diagnosis ($p = 0.08$), primary versus middle/high school attendance ($p = 0.06$), and neurocognitive disorders at baseline ($p = 0.08$).

Discussion

At 1 month, mean CPAP adherence was high (6.1 h/night) and did not decrease during the 24-month follow-up. High compliance (8.4 h/night) and a correction of OSA indices at a mean of 12.3-month follow-up have already been reported by Amaddeo et al. [8] in 31 children initiated in an outpatient setting. However, their studied population from a tertiary highly specialized center was heterogeneous including syndromic and non-syndromic children. Our data suggest that initiating CPAP in an ambulatory mode in non-syndromic OSA children is safe, acceptable, and seemingly effective in the long term, reflecting tolerance and perceived benefits by children and their families.

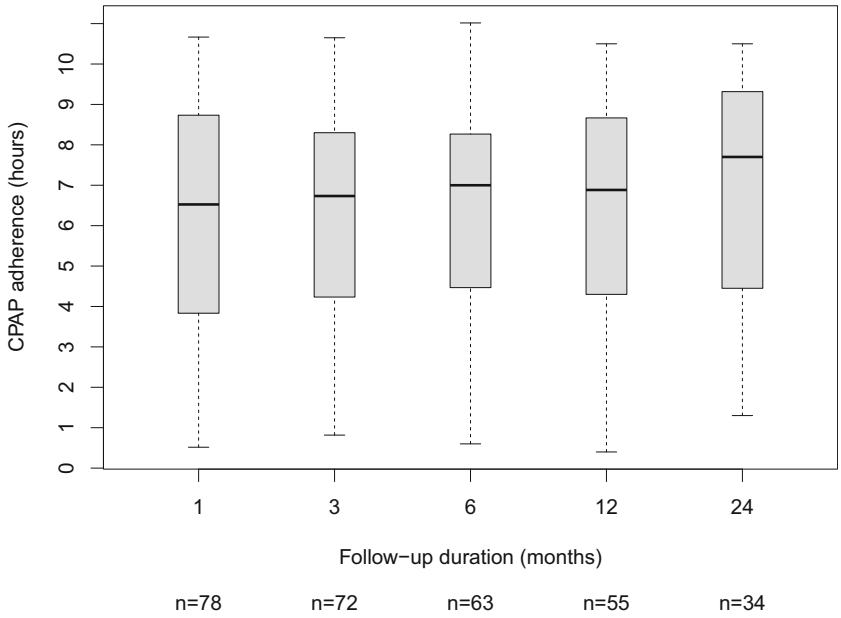

Fig. 2 Evolution of CPAP adherence during the 24-month follow-up

The significant percentage of children found to have been cured by an alternative strategy to CPAP at the annual polysomnographic reassessments justifies periodic reevaluation for adjustment of the management plan. Our study results conflict with the poor adherence reported by Hawkins et al. [9], in which only 49% of children were adherent (> 4 h/night). That study was conducted in the USA without home care providers performing home visits. CPAP adherence was assessed as the mean number of hours per night because in France, data on "nights per week" CPAP usage are not mandatory for reimbursement and were not available in our dataset.

For many of our secondary endpoints, we only observed trends, and we acknowledge that the study might lack power to achieve statistical significance. The trend we observed towards an association between OSA severity and CPAP adherence is in contradiction to Hawkin's retrospective study [9], which reported that OSA (diagnostic AHI and degree of hypoxemia), therapeutic pressure, and residual AHI did not impact CPAP adherence. The trend we found between age and CPAP adherence is in line with the results of DiFeo et al. [10], who suggested that educational programs for pediatric patients and their families should differ with age range in order to improve CPAP adherence [11], especially in adolescents with OSA who need to be better supported in their use of CPAP therapy [12]. The good CPAP adherence demonstrated by our cohort is expected to be associated with improvements in neurocognition, quality of life [13], and metabolic and cardiovascular parameters. Specific integrated care support at home is certainly an important factor in improving self-efficacy when starting CPAP therapy in children and young people with OSA [14].

To conclude, ambulatory CPAP initiation and follow-up is less time-consuming and likely to be cost-effective for children and their families. The effectiveness of alternatives to CPAP (rehabilitation programs, dento-facial orthodontics, and weight loss) is significant. CPAP is never initiated as a single therapy, and a reevaluation by polysomnography is needed before pursuing or stopping CPAP as the child grows up.

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Compliance with ethical standards

Conflict of interest The authors declare that they have no conflict of interest.

Ethical approval All procedures performed in studies involving human participants were in accordance with the ethical standards of the institutional and/or national research committee and with the 1964 Helsinki declaration and its later amendments or comparable ethical standards.

Informed consent Informed consent was obtained from both parents of all individual participants included in the study.

References

1. Kaditis AG, Alonso Alvarez ML, Boudewyns A, Alexopoulos EI, Ersu R, Joosten K, Larramona H, Miano S, Narang I, Trang H, Tsaoussoglou M, Vandenbussche N, Villa MP, Van Waardenburg D, Weber S, Verhulst S (2016) Obstructive sleep disordered breathing in 2- to 18-year-old children: diagnosis and management. Eur Respir J 47:69–94
2. Sundaram SS, Halbower AC, Klawitter J, Pan Z, Robbins K, Capocelli KE, Sokol RJ (2018) Treating obstructive sleep apnea and chronic intermittent hypoxia improves the severity of nonalcoholic fatty liver disease in children. J Pediatr 198:67–75.e1
3. DelRosso LM, King J, Ferri R (2018) Systolic blood pressure elevation in children with obstructive sleep apnea is improved with positive airway pressure use. J Pediatr 195:102–107.e1
4. Uong EC, Epperson M, Bathon SA, Jeffe DB (2007) Adherence to nasal positive airway pressure therapy among school-aged children and adolescents with obstructive sleep apnea syndrome. Pediatrics 120:e1203–e1211
5. Marcus CL, Radcliffe J, Konstantinopoulou S, Beck SE, Cornaglia MA, Traylor J, DiFeo N, Karamessinis LR, Gallagher PR, Meltzer LJ (2012) Effects of positive airway pressure therapy on neurobehavioral outcomes in children with obstructive sleep apnea. Am J Respir Crit Care Med 185:998–1003
6. Ramirez A, Khirani S, Aloui S, Delord V, Borel J-C, Pépin J-L, Fauroux B (2013) Continuous positive airway pressure and noninvasive ventilation adherence in children. Sleep Med 14:1290–1294
7. Marcus CL, Rosen G, Ward SLD, Halbower AC, Sterni L, Lutz J, Stading PJ, Bolduc D, Gordon N (2006) Adherence to and effectiveness of positive airway pressure therapy in children with obstructive sleep apnea. Pediatrics 117:e442–e451
8. Amaddeo A, Frapin A, Touil S, Khirani S, Griffon L, Fauroux B (2018) Outpatient initiation of long-term continuous positive airway pressure in children. Pediatr Pulmonol 53:1422–1428
9. Hawkins SMM, Jensen EL, Simon SL, Friedman NR (2016) Correlates of pediatric CPAP adherence. J. Clin. Sleep Med. JCSM Off. Publ. Am. Acad. Sleep Med 12:879–884
10. DiFeo N, Meltzer LJ, Beck SE, Karamessinis LR, Cornaglia MA, Traylor J, Samuel J, Gallagher PR, Radcliffe J, Beris H, Menello MK, Marcus CL (2012) Predictors of positive airway pressure therapy adherence in children: a prospective study. J. Clin. Sleep Med. JCSM Off. Publ. Aem. Acad. Sleep Med 8:279–286
11. Jambhekar SK, Com G, Tang X, Pruss KK, Jackson R, Bower C, Carroll JL, Ward W (2013) Role of a respiratory therapist in improving adherence to positive airway pressure treatment in a pediatric sleep apnea clinic. Respir Care 58:2038–2044
12. Alebraheem Z, Toulany A, Baker A, Christian J, Narang I (2018) Facilitators and barriers to positive airway pressure adherence for adolescents. A qualitative study. Ann Am Thorac Soc 15:83–88
13. Lynch MK, Elliott LC, Avis KT, Schwebel DC, Goodin BR (2017) Quality of life in youth with obstructive sleep apnea syndrome (OSAS) treated with continuous positive airway pressure (CPAP) therapy. Behav Sleep Med 30:1–8
14. Xanthopoulos MS, Kim JY, Blechner M, Chang MY, Menello MK, Brown C, Matthews E, Weaver TE, Shults J, Marcus CL (2017) Self-efficacy and short-term adherence to continuous positive airway pressure treatment in children. Sleep 40(7). https://doi.org/10.1093/sleep/zsx096