pii: jc-18-00362 https://dx.doi.org/10.5664/jcsm.7850

JCSM
Journal of Clinical Sleep Medicine

SCIENTIFIC INVESTIGATIONS

Adherence to Positive Airway Pressure for the Treatment of Obstructive Sleep Apnea in Children With Developmental Disabilities

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Study Objectives: To determine whether adherence to positive airway pressure (PAP) differs in children with developmental disabilities (DD) compared to typically developing (TD) children.

Methods: PAP adherence of 240 children initiated on PAP for obstructive sleep apnea (OSA) was retrospectively analyzed. Adherence between groups, expressed as percentage of nights used and hours of usage on nights used at 3 and 6 months, was compared. Predictive factors of adherence were studied using a median regression model.

Results: A total of 103 children with DD (median [interquartile range] age = 7.9 [3.2–13.1] years) and 137 TD (11.0 [5.5–16.1], *P* = .005) children were included. Percentage of nights used was significantly higher in children with DD at 3 (DD = 86.7 [33.9–97.9], TD = 62.9 [30.8–87.8] *P* = .01) and 6 months (DD = 90.0 [53.3–100], TD = 70.7 [29.2–90.8], *P* = .003). Hours of usage on nights used at 3 and 6 months were similar between groups (DD = 5.0 [1.4–7.9], TD = 4.6 [1.9–7.2], *P* = .715; DD = 6.4 [1.8–8.3], TD = 5.7 [2.5–7.3], *P* = .345, respectively). This adherence measure improved over time in both groups (DD, *P* = .007; TD, *P* = .005). At 6 months, higher median neighborhood income and titration at or before 6 months were significantly predictive for percentage of nights used; higher PAP pressure was significantly predictive for hours of usage in both groups.

Conclusions: Children with DD had better PAP adherence expressed as percentage of nights used than TD children. Hours of usage on nights used at 3 and 6 months were similar between groups and improved over time. Higher income and titration at or before 6 months were predictive of adherence in all children. These findings indicate that children with DD can successfully wear PAP.

Keywords: adherence, children, developmental disabilities, obstructive sleep apnea, positive airway pressure

Citation: Kang EK, Xanthopoulos MS, Kim JY, Arevalo C, Shults J, Beck SE, Marcus CL, Tapia IE. Adherence to positive airway pressure for the treatment of obstructive sleep apnea in children with developmental disabilities. *J Clin Sleep Med.* 2019;15(6):915–921.

BRIEF SUMMARY

Current Knowledge/Study Rationale: Children with developmental disabilities (DD) requiring positive airway pressure (PAP) for the treatment of obstructive sleep apnea are a growing and understudied population. Research has shown that children with DD could benefit from behavioral interventions to improve PAP adherence, but few studies have specifically analyzed PAP adherence in this group.

Study Impact: This study showed that children with DD, with adequate support, have better PAP adherence than typically developing children. The study also identified predictive factors of PAP adherence in children.

INTRODUCTION

Positive airway pressure (PAP) is the second-line treatment for the obstructive sleep apnea (OSA) in children.[1] PAP has been shown to be highly effective in maintaining airway patency and improving neurobehavioral outcomes.[2,3] However, it has also been demonstrated that the improvement in neurobehavioral outcomes correlates with adherence. Specifically, children who wear PAP consistently and for longer periods of time overnight benefit the most of this therapy.[2] This may be particularly important for vulnerable populations, such as children with developmental disabilities (DD). Further, several DD are characterized by a higher prevalence of OSA. For example, OSA prevalence in children with Down syndrome has been reported to range from 30%[4] to 66.4%,[5] and OSA prevalence in children with Prader-Willi has been estimated at 79.9%.[6] Nevertheless, there are limited data specifically describing PAP adherence in children with DD as previous studies have grouped together typically developing (TD) children and those with DD. Therefore, children who have both DD and OSA merit further research.

Children with DD may face similar PAP implementation issues as TD children, such as incorporating PAP into their routine, getting used to the sensation of the mask, headgear, and pressure, as well as the need for caregiver support. However, they may also encounter difficulties related to their disability that may deter caregivers and healthcare providers to pursue PAP, such as sensory and behavioral concerns. Nonetheless, similar PAP adherence in adult participants with and without intellectual disabilities has been reported

following an intensive inpatient protocol.[7] However, such a protocol may not be feasible in many healthcare facilities and may also not be feasible for children and families. We conducted a retrospective analysis comparing PAP adherence for the treatment of OSA over a 6-month period in children with DD and TD children initiated on PAP by an interdisciplinary team in an outpatient setting as described previously.[8] Briefly, children are evaluated by a pediatric sleep physician, a pediatric behavioral psychologist, a sleep registered nurse, a respiratory therapist, and occasionally a social worker. Based on published correlates of PAP adherence,[9] we hypothesized that children with DD would have better adherence than TD children.

METHODS

Study Group

This is a retrospective study that included children aged 0 to 18 years initiated on PAP for the treatment of OSA between January 1, 2015 and December 31, 2016. Children initiated on PAP for conditions other than OSA (eg, respiratory failure) were excluded. Two investigators (EK and CA) reviewed charts independently, before extracting PAP adherence data, to determine whether children were typically developing or had developmental disabilities. Developmental disabilities included genetic syndromes (eg, Trisomy 21, fragile X, Rett's, Prader-Willi), central nervous system abnormalities (eg, congenital cytomegalovirus infection, stroke, hydrocephalus, cerebral palsy), autism, and idiopathic causes. Final agreement on developmental status was reached through discussion.

Data Collection

Medical records were reviewed to extract age, date of PAP initiation, gender, race, ZIP codes, body mass index (BMI) z-score, diagnosis (cause of OSA), comorbidities, presence of developmental disability (yes/no), baseline polysomnography results, and PAP titration date. PAP adherence data at 1, 3 and 6 months were collected using Encoreanywhere software (Respironics, Murrysville, Pennsylvania, United States) when available, or manually using adherence data downloaded from the data card. Adherence was expressed as percentage of nights used and hours of usage on nights used, at 3 and 6 months from PAP initiation. PAP mode (bilevel [BPAP], continuous [CPAP], auto CPAP) and PAP pressure were also evaluated. In the case of BPAP, the pressure was calculated as the average of inspiratory PAP and expiratory PAP. In the case of auto PAP, the auto PAP peak pressure was utilized for analyses. The dropouts were defined as children who did not return for follow-up and whose devices were not able to be downloaded via modem. Median neighborhood income was generated by entering ZIP codes into the American Community Survey.[10] Median income for international addresses could not be reliably obtained. Total apnea-hypopnea index (AHI) was defined as the sum of obstructive and central apneas, obstructive and central hypopneas, and mixed apneas divided by the total sleep time. The study protocol was approved by Children's Hospital of Philadelphia Institutional Review Board.

Data Analysis

Baseline and demographic characteristics were summarized by standard descriptive statistics (eg, median and interquartile range for quantitative variables such as age and percentages for categorical variables such as sex). PAP usage was compared between children with DD and TD children using a Wilcoxon rank-sum test, as data were not normally distributed. PAP predictive factors for adherence at 6 months were evaluated by median regression. DD, race, sex, PAP pressure, titration status at 6 months, median neighborhood income quartiles, obesity and the baseline obstructive apnea-hypopnea index (OAHI) were included in this model. OAHI was defined as the sum of obstructive apneas, obstructive hypopneas and mixed apneas divided by the total sleep time. The interaction term between PAP pressure and titration status was considered. Changes in adherence over time were analyzed using the linear quantile mixed model, which is particularly useful when dealing with repeated measures. A P value < .05 was considered significant. Analyses were performed with R version 3.4.0 (2017-04-21).

RESULTS

Study Group Characteristics

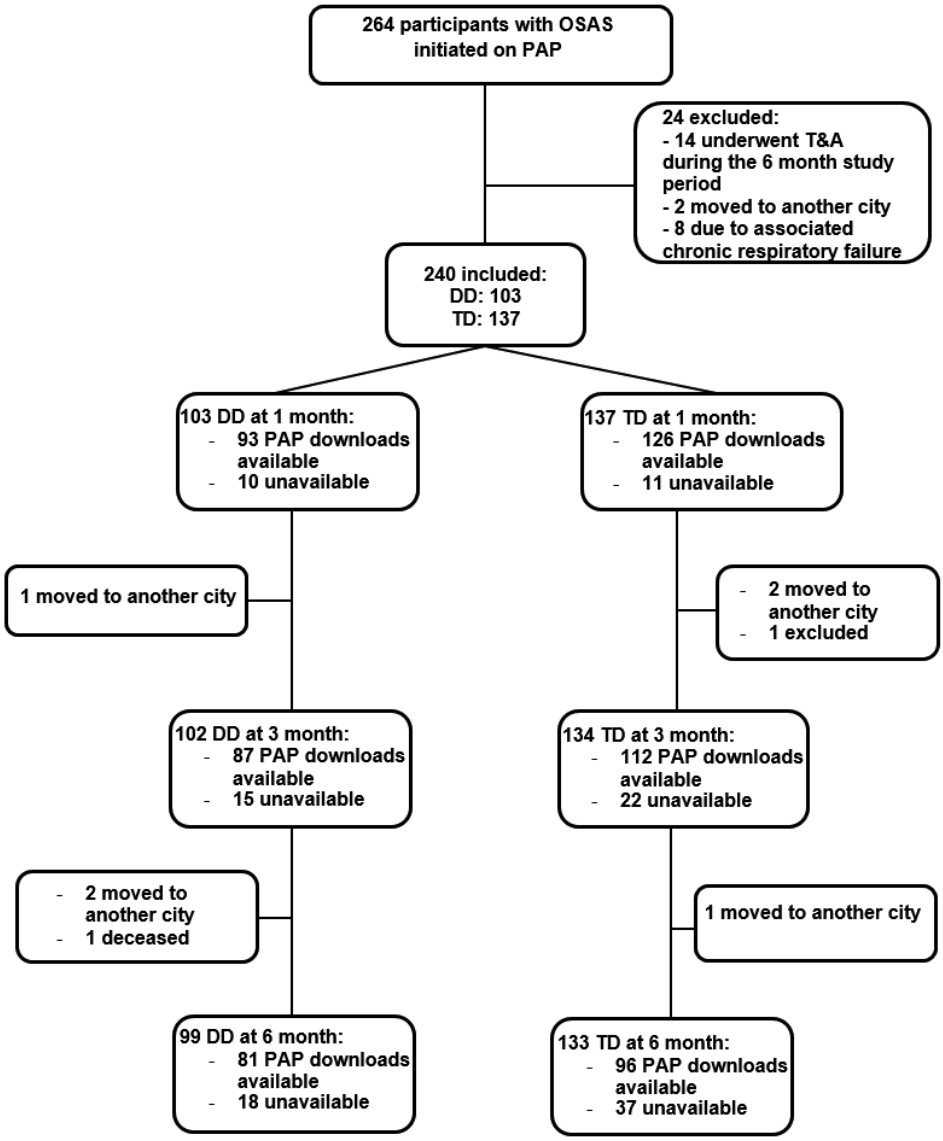
A total of 264 participants were initiated on PAP during the study period as shown in **Figure 1**. Of these, 24 were excluded due to chronic respiratory failure or adenotonsillectomy during the 6-month period following initiation. A total of 240 children were included in analyses. The participants' characteristics are shown in **Table 1**. Children with DD lived in more affluent neighborhoods; were younger, leaner, and predominantly Caucasian. Median income data of 5 children with international addresses were not obtained. Genetic syndrome was the most common cause of DD and Down syndrome was the prominent genetic disorder (31% of all children with DD) as shown in **Table 2**.

Adherence in Children With DD and TD Children

PAP adherence expressed as percentage of nights used at 3 and 6 months was significantly higher in children with DD (**Figure 2**). Specifically at 3 months, adherence was 86.7% (33.9% to 97.9%) in children with DD and 62.9% (30.8% to 87.8%) in TD children (P = .01). At 6 months, adherence in children with DD was 90.0% (53.3% to 100%) and in TD children was 70.7% (29.2% to 90.8%) (P = .003). Hours of usage on nights used at 3 and 6 months were similar between both groups (P = .715 at 3 months, P = .345 at 6 months, **Figure 3**). Importantly, and according to the linear quantile mixed model for each group for each adherence variable, this adherence measure also improved at 6 months compared with that at 1 month in both groups (DD P = .007; TD P = .005). The overall median nightly use was 5.72 hours at 6 months.

There were 55 dropouts (18 in DD, 37 in TD group, **Figure 1**) in the adherence data at 6 month. The missing rate was similar between DD and TD groups (17.5% versus 23.4%, respectively,

Figure 1—Flow diagram of participants.



DD = developmental disabilities, OSA = obstructive sleep apnea, PAP = positive airway pressure, T&A = adenotonsillectomy, TD = typically developing.

P = .118). As shown in **Table 3**, missing and non-missing participants were similar except for titration study performed at or before 6 months and PAP pressure that were significantly higher in those with complete data.

Adherence Factors

Adherence factors at 6 months of initiation were analyzed. 66% of children had a titration sleep study performed at or before this time point. Therefore, an interaction term between PAP pressure and titration status was included in the regression model to assess for possible differences between children receiving polysomnography-certified therapeutic pressures versus those still receiving empiric pressures. The interaction was not significant. For all children regardless of developmental status, higher income quartiles (second quartile P = .002, third quartile P = .001, fourth quartile P = .007) and titration performed at or before 6 months of initiation (P = .012) were identified as significant predictive factors for percentage of nights used. Higher PAP pressure was a significant predictive factor for hours of usage on nights used.

DISCUSSION

This study reveals that children with DD had better PAP adherence expressed as percentage of nights used than TD children at 3 and 6 months. The hours of usage on nights used at 3 and 6 months were similar between both groups. However, and unlike previous reports,[11,12] this adherence measure improved over time in both groups. Results also identified higher median neighborhood income and higher PAP pressure as predictors of PAP usage in the entire group. These findings suggest that children with DD, with adequate support, can successfully wear PAP.

Table 1—Demographic characteristics of the study group.

Characteristics	DD (n = 103)	TD (n = 137)	*P*
Age at PAP (years)	7.9 (3.2–13.1)	11.0 (5.5–16.0)	.005
Male, n (%)	59 (57.38)	82 (59.8)	.694
Race, n (%)			< .001
African American	25 (24.3)	79 (57.7)	
Caucasian	53 (51.5)	33 (24.1)	
Other	25 (24.3)	25 (18.2)	
Annual neighborhood income ($, in thousands)	67.4 (43.0–86.6)	47.8 (30.8–73.3)	.0015
BMI z-score	1.53 (0.71–2.13)	2.26 (0.80–2.73)	.001
Weight/height percentile (n = 24)	78.8 (30.7–90.9)	86.2 (55.0–95.7)	.371
Obese, n (%)	43 (40.8)	85 (62.0)	.002
Polysomnography			
OAHI (events/h)	16.4 (9.1–26.7)	11.8 (9.0–27.6)	.199
Total AHI (events/h)	18 (10.9–51.8)	12.9 (9.4–18.5)	.055
SpO_2 nadir (%)	83.0 (76.0–88.2)	86.0 (79.0–89.0)	.026
PAP pressure (cmH_2O)	8.0 (5.0–9.8)	6.0 (5.0–8.0)	.065

Data are presented as median (interquartile range) or n (%). *P* values for numeric variables were derived from Wilcoxon rank sum test. *P* values for categorical variables were derived from the Fisher exact test. BMI z-score was provided for those age ≥ 2 years, and weight/height percentile for those < 2 years. AHI = apnea-hypopnea index, OAHI = obstructive apnea-hypopnea index, PAP = positive airway pressure, SpO_2 = oxyhemoglobin saturation.

Table 2—Diagnosis associated with developmental disabilities (n = 103, non-overlapping).

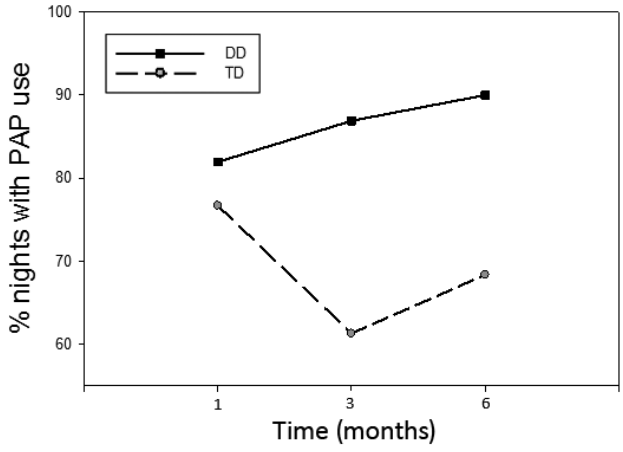
Genetic Syndrome (n = 48, 46.6%)	Trisomy 21 (32), Prader-Willi syndrome (4), DiGeorge syndrome (3), Trisomy 18 (1), Soto syndrome (1), fragile X syndrome (1), Smith-Magenis syndrome (1), Coffin-Siris syndrome (1), Lowe syndrome (1), Wolf-Hirschhorn syndrome (1), chromosome 4Q deletion (1), other chromosome abnormality (1)
CNS Abnormalities (n = 24, 23.3%)	Cerebral palsy (11), Arnold-Chiari malformation/Spina bifida (6), hydrocephalus (3), Dandy Walker malformation (1), empty sella syndrome (1), septo-optic dysplasia (1), agenesis of corpus callosum (1), periventricular leukomalacia (1)
Autism Spectrum Disorder (n = 12, 11.6%)	(including Rett syndrome, Asperger syndrome)
Other (n = 19, 18.4%)	Idiopathic/global developmental delay (10), epilepsy (3), craniosynostosis (2), mucolipidosis type II (1), unspecified intellectual disability (1), Renpenning syndrome (1), neurofibromatosis type 1 (1)

Most studies in children, have reported suboptimal PAP adherence with a mean nightly use of 3.4–5.3 hours.[13–15] However, some studies have reported higher use.[8] In the current study, overall median nightly use was 5.72 hours at month 6 (6.38 hours in DD, 5.65 hours in TD group). These findings show that regardless of developmental ability, PAP adherence in children can be successful. Longer-term data are necessary to determine if the trajectory of increased use persists after 6 months. A strength of the study is the clearly characterized groups and evaluation of the trajectory of use over time.

There are limited data regarding PAP adherence in children with DD. A previous study in a sample of 56 children of whom 23% had DD, reported that PAP adherence was not affected by DD.[16] Brooks et al studied 25 children with Down syndrome to analyze the relationship between sleep and cognition, and showed that of the 7 children who were treated with PAP only 3 were able to use it at home.[17] Another recent study reported that children with DD were very likely to be adherent to CPAP with an odds ratio of 2.55 ($P = .007$).[9] Authors attributed these results to increased dependence on caregiver support, increased parental perception of PAP necessity, and decreased ability of the patient to remove the interface. However, children with Down syndrome were analyzed as a third independent group that did not appear to influence adherence. The current study adds to the literature by comparing a large group of children with DD to those without DD. One recent small study in adults with intellectual disabilities requiring PAP described an intensive inpatient protocol to train participants and caregivers.[7] Data showed that PAP adherence in adults with intellectual disabilities 6 months after initiation was comparable to that of controls. Specifically, 65% of adults with intellectual disabilities used PAP at least 70% of the nights and more than 4 hours per night compared to 50% of adults without intellectual disabilities. The authors hypothesized that this intensive initial support could influence long-term adherence. While this study is promising, the feasibility and generalizability of an intensive inpatient protocol is debatable given the high cost and limited resources of inpatient care at most institutions.

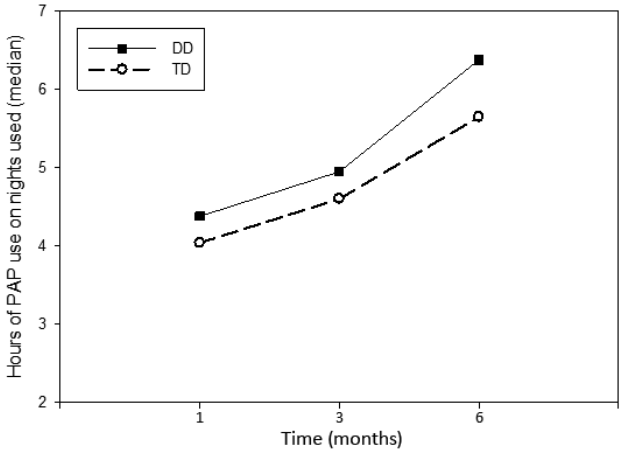
PAP adherence appears to be multifactorial. However, predictive factors have not been consistently identified across all studies.[18] This may be partly due to the heterogeneity of the population requiring PAP and to the retrospective nature

Figure 2—Adherence over 6 months to PAP in DD and TD groups, expressed as % of nights used.



Children in the DD group had better PAP adherence at 6 months, expressed as % of nights used, than those in the TD group (P = .003). DD = developmental disabilities, PAP = positive airway pressure, TD = typically developing.

Figure 3—Adherence over 6 months to PAP in DD and TD groups, expressed as median daily use in hours.



Adherence expressed as hours of PAP used on the nights used significantly increased over time in both groups (DD P = .007; TD P = .005). However, this measure was similar between both groups (at 6 months, P = .345). DD = developmental disabilities, PAP = positive airway pressure, TD = typically developing.

Table 3—Comparison of demographic characteristics between the those with PAP download available at 6 months and dropouts.

Variables	Dropouts (n = 55)	PAP Download Available (n = 177)	*P*
Age at PAP (years)	7.2 (2.8–14.4)	10.6 (5.5–14.7)	.126
Male, n (%)	30 (54.5)	106 (59.9)	.532
Race, n (%)			.762
African American	21 (38.2)	79 (44.6)	
Caucasian	21 (38.2)	63 (35.6)	
Other	13 (23.6)	35 (19.8)	
Annual neighborhood income ($, in thousands)	57.9 (41.8–72.9)	52.3 (30.8–84.4)	.546
BMI z-score *	1.56 (0.89–2.61)	1.87 (0.72–2.53)	.963
Weight/height percentile (n = 12) *	81.3 (21.1–87.6)	85.2 (56.6–96.1)	.522
Obese, n (%) *	24 (43.6)	94 (53.1)	.175
Polysomnography			
OAHI (events/h)	11.2 (8.1–22.1)	14.7 (9.5–29.0)	.070
Total AHI (events/h)	13 (9.3–23.9)	15.8 (9.8–30.5)	.186
SpO_2 nadir (%)	85.0 (79.0–90.0)	84.0 (77.0–89.0)	.265
PAP pressure (cmH_2O)	5.0 (4.0–7.0)	7.8 (5.0–9.0)	< .001
Developmental disability, n (%)	18 (32.7)	81 (45.8)	.118
Titration at or before 6 months, n (%)	25 (39.9)	134 (75.7)	< .00001

Values are presented as median (interquartile range) or n (%). P values for numeric variables were derived from Wilcoxon rank sum test. P values for categorical variables were derived from the Fisher exact test. BMI z-score was provided for those age ≥ 2 years, and weight/height percentile for those < 2 years. AHI = apnea-hypopnea index, OAHI = obstructive apnea-hypopnea index, PAP = positive airway pressure, SpO_2 = oxyhemoglobin saturation.

of studies. In one review, factors consistently associated with nonadherence to PAP in adults and children included being asymptomatic, nasal obstruction, low self-efficacy, lack of risk perception and lower socioeconomic status.[19] In children, maternal education has been identified as the strongest predictor of PAP adherence, and African American race has been associated with lower adherence.[16] However, OSA severity and PAP pressure have not been found to influence adherence. The current findings support previous research that higher socioeconomic status, here expressed as median neighborhood income, is associated with greater adherence; whereas they differ in that higher PAP pressure was identified as a predictor of adherence for the entire group. A plausible explanation for this may rely on the fact that children with DD were more adherent and lived in more affluent neighborhoods. Higher PAP pressures resulted in better usage. Possibly, children with higher

PAP may have more severe OSA and therefore, families might be more mobilized to treat the OSA. It is also possible that individuals with more severe OSA experienced greater feelings of benefit or improvement from using PAP (eg, less sleepy, easier to wake in the morning, less irritability). Similarly, dropouts had lower pressures and borderline lower OAHI. It is possible that dropouts had stopped using PAP due to lack of perceived benefits. Importantly, according to the statistical model, the presence of a titration study performed at or before 6 months did not appear to influence these results. Data of secondary improvements were not collected in this study and future prospective studies should include these patient-centered outcomes.

There are no consistent findings in the literature regarding PAP modality influence on adherence. An Australian pediatric cohort study showed that adherence to BPAP was greater than for CPAP.[20] However, the BPAP group included more subjects with neuromuscular diseases than the CPAP group (36.4 versus 5.5%, respectively). Therefore, results may have been confounded by respiratory insufficiency dependency on noninvasive ventilation. Previous results from our group showed no difference in adherence between BPAP and CPAP users in children.[21] Other previously reported PAP adherence factors in children include type of interface, time from initiation to children's initial acceptance of PAP, higher self-reported quality of life and lower BMI.[18] Disease severity, sex, previous upper airway surgery, concomitant psychological support with PAP initiation, and mode of PAP delivery were not associated with PAP use.[18] In another smaller study of PAP adherence among school-aged children and adolescents following adenotonsillectomy, adherence was higher in those with higher baseline OAHI.[22] In the current study, baseline OAHI was not associated with adherence.

In the present study, adherence over the 6-month observation period increased linearly in the DD group, but the TD group displayed a different trajectory. The TD group's adherence decreased from month 1 to month 3 and then increased from month 3 to month 6. These results suggest that families of TD children may be more motivated in the first month of treatment, potentially due to the novelty and recent intensive intervention at the first visit, but then adherence wanes and there is a window of opportunity to increase adherence between 3 and 6 months in TD children. To the best of our knowledge, there are no previous pediatric data reporting adherence trajectory. Another notable finding was that TD children used PAP for less nights compared to children with DD, but usage hours on nights used was similar. One plausible explanation for this finding might be that TD children might have more autonomy in PAP usage and take a night off, eg, weekend or a sleep over, a common finding in our practice.

There are some limitations in this study. Due to the retrospective nature, some important variables such as maternal education, family income, interface use/preference, and patient-reported outcomes were not available. The study period was somewhat short and longer-term studies beyond 6 months are needed. A total of 63 participants met drop out criteria during the 6-month study period. This is similar to other investigations.[9]

In summary, children with DD had better adherence to PAP expressed as percentage of nights used compared to TD children and showed increasing adherence over time over 6 months. The hours of usage on nights used at 3 and 6 months were similar between both groups and, importantly, improved over time in both groups. Higher median neighborhood income and higher PAP pressure were identified as predictive factors in children with and without DD. These findings show that children with DD, with adequate support, can successfully initiate and wear PAP. Studying a prospective cohort of children with DD and TD would help to further elucidate predictors of adherence and risk factors for nonadherence.

ABBREVIATIONS

AHI, apnea-hypopnea index
BMI, body mass index
BPAP, bilevel positive airway pressure
CPAP, continuous positive airway pressure
DD, developmental disabilities
OAHI, obstructive apnea-hypopnea index
OSA, obstructive sleep apnea
PAP, positive airway pressure
TD, typically developing

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ACKNOWLEDGMENTS

The authors thank the sleep nurses, technicians, and physicians; respiratory therapists; and children and their families.

SUBMISSION & CORRESPONDENCE INFORMATION

Submitted for publication June 14, 2018
Submitted in final revised form October 16, 2018
Accepted for publication November 19, 2018
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DISCLOSURE STATEMENT

Work for this study was performed at Children's Hospital of Philadelphia, Philadelphia, PA. All authors have seen and approved this manuscript. The authors report no conflicts of interest. Support: NIH UL1RR024134, K01HL130719, and Research Electronic Data Capture (REDCap).