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ORIGINAL RESEARCH

To Wear or Not Wear the Mask: Decline in Positive Airway Pressure Usage in Children with Sleep Disordered Breathing During the COVID-19 Pandemic

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Purpose: Positive airway pressure (PAP) therapy is an effective treatment prescribed to children with sleep disordered breathing (SDB); however, PAP adherence remains challenging. Given that COVID-19 pandemic continues to impact sleep and daily life, the aim of this study was to evaluate longitudinal trajectory of PAP usage in children during the COVID-19 pandemic.

Patients and Methods: This was a retrospective study. Children aged 1–18 years with SDB prescribed PAP at The Hospital for Sick Children (Toronto, Canada) were evaluated for PAP adherence. Demographics, medical history and PAP adherence data during four consecutive 3-month time periods from December 2019 to December 2020 were collected. These four time periods included i) prior to COVID-19 lockdown, ii) during the first three months of lockdown, iii) summer and iv) return to school period. Percentage of days where PAP was used for ≥4 hours and average nightly usage of PAP were primary outcomes.

Results: A total of 149 children (61.7% male, mean (±SD) age=12.8 ± 4.1 years, BMI (±SD) z-score=1.45±1.43) were enrolled. Compared to prior to lockdown, the median (IQR) of percentage of PAP usage ≥4 hours and average nightly usage of PAP declined significantly during the summer and return to school periods ($p<0.001$ for all). By the end of the return to school period, only 69/149 (46%) showed sustained PAP usage and 80/149 (54%) had decreased PAP usage. Obesity was a risk factor for a decline in PAP usage after returning to school ($\beta=-15.36$, $p=0.03$).

Conclusion: Compared to COVID-19 pre-pandemic PAP usage, there was a significant decline in PAP usage across COVID-19 pandemic. There is critical under usage of PAP in children diagnosed with SDB, resulting in an urgent need to address barriers to mitigate poor adherence to PAP long-term. Targeted strategies are required to optimize PAP adherence in children with SDB.

Keywords: COVID-19 pandemic, sleep disordered breathing, longitudinal, positive airway pressure adherence, children

Introduction

Positive airway pressure (PAP) therapy such as continuous positive airway pressure (CPAP) and bi-level positive airway pressure (Bi-level PAP) is commonly prescribed to children in the home with chronic respiratory disorders including sleep disordered breathing (SDB), chronic lung disease and neuromuscular weakness.[1,2] SDB encompasses obstructive sleep apnea (OSA), central sleep apnea (CSA) and/or nocturnal hypoventilation (NH).[3,4] Consistent PAP use is highly efficacious for eliminating the nightly SDB and associated with several health benefits including better mood, improvements in neurocognitive and behavioral function, enhanced academic performance, improved quality of life and reductions in cardio-metabolic risk in children.[4–6] Although PAP is an effective treatment for SDB, adherence remains

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a significant challenge with less than 50% of children using PAP more than 4 hours a night, thereby reducing the effectiveness of PAP.[7–11] Interestingly, these observations are reported across all ages and developmental stages in children.[9–14] Factors positively influencing PAP adherence include underlying medical diagnosis (children with developmental delay were more likely to be adherent with PAP than those without a developmental delay diagnosis), practical caregiver support, patient engagement programs, time since PAP was initially prescribed, having a family member on PAP therapy as a "role model", physical design of the PAP machine and perceiving benefits of PAP use.[11,14–16]

Given the challenges of PAP adherence, there are concerns regarding the impact of the COVID-19 pandemic as a result of severe acute respiratory syndrome coronavirus 2 (SARS-CoV-2) on PAP adherence.[17] It is now well documented that access and delivery of medical care has changed dramatically from in-person office visits to virtual care as a result of COVID-19 pandemic.[18] Specifically, within sleep medicine, COVID-19 pandemic has impacted access to polysomnograms (PSG) eg, reduction of available in laboratory PSG and reduction in home sleep testing; pivoted in-person clinic visits to virtual visits and delays in receiving therapeutic interventions.[19,20] Interestingly, a study of 7485 adults in France showed that CPAP adherence improved during a one-month period of the COVID-19 lockdown compared to one-month prior to the lockdown from an average of 386 minutes per night prior to the lockdown to 401 minutes per night during lockdown, resulting in a 3.9% increase in adherence.[21] A cross-sectional study of CPAP adherence in 101 adults with OSA showed that CPAP adherence significantly improved during a 40-day period of the COVID-19 lockdown compared to a 6-month period prior to the lockdown.[22] Factors associated with improved PAP adherence included good CPAP adherers prior to lockdown, the female gender and younger age.[22] Similarly, in our own center, we recently published data on 151 children with SDB prescribed PAP therapy with objective data showing that 63% of children with SDB in fact had increased PAP adherence during the first 3 months of lockdown compared to 3 months prior to lockdown; specifically, 1) an increase in the median of percentage of days of PAP usage for $\geq$ 4 hours (76.7 versus 62.0%, p = 0.02) and 2) average nightly PAP usage (406.0 versus 367.0 minutes, p = 0.006) respectively.[23] However, there is emerging data that highlights significant changes in sleep schedule, physical activity behaviours and mental health of children and caregivers.,[24,25] which together may adversely affect longer-term pediatric PAP adherence. We hypothesized that despite the increase in PAP adherence observed in our cohort immediately during the lockdown period,[23] PAP adherence would subsequently decrease over time to usage observed prior to the COVID-19 pandemic. Thus, the aim of this study was to evaluate and compare PAP adherence at four distinct 3-month time periods which included prior to COVID-19 lockdown, during the first three months of lockdown, summer period (no school) and return to school.

Materials and Methods

Study Population

Children aged 1–18 years old with a diagnosis of OSA, CSA and/or NH prescribed PAP at The Hospital for Sick Children (Toronto, Canada) were eligible for this study. Specifically, the study population consisted of all children who were prescribed PAP at home prior to the COVID-19 pandemic in Ontario, Canada and continued to be prescribed PAP over the subsequent year. We did not deselect or exclude any patients based on their PAP usage at the time. Adherence to PAP was evaluated performed during four consecutive 3-month time periods as follows: 1) prior to COVID-19 pandemic in Ontario, Canada (December 18, 2019- March 16, 2020); 2) during the first three months of the pandemic (March 17-June 14, 2020), with the COVID-19 lockdown, declared on March 17th, 2020 in Ontario, Canada; 3) summer (June 15-September 13, 2020) and 4) return to school (September 14- December 12, 2020). Children were excluded if 1) PAP adherence download data was not available during all four distinct time periods and 2) Children were asked to terminate PAP by their medical provider during this time period. Objective PAP usage was obtained via PAP device data reports acquired remotely. Medical records were retrospectively reviewed which included baseline demographics (age, gender, height, weight and body mass index (BMI)), medical history such as primary diagnosis, comorbidities, allergies, medications, type of SDB, history of adenotonsillectomy (AT), type of PAP therapy, duration of initiation of PAP therapy, independent PAP user (eg able to apply and use PAP at home independently of parental/caregiver support as

determined by the respiratory therapist following the child's PAP adherence patterns), and PAP adherence data. Obesity was characterized by a BMI more than or equal to the 95th percentile for gender and age.[26]

PAP Adherence

PAP usage was assessed remotely via device data reports as previously reported.[23] All participants used ResMed PAP devices and objective PAP adherence were assessed by the software including Airview for CPAP (ResMed; San Diego, CA, USA) and ResScan for Bi-level PAP (ResMed; San Diego, CA, USA).[27]

The primary outcomes were i) percentage of days where PAP was used for 4 hours or more usage (%) and ii) average nightly usage (minutes) defined as the average minutes of PAP use based on only the days where PAP was used. Overall percentage of days that PAP was used (regardless of duration of usage), and average total usage (minutes) which was defined as the average minutes of PAP use based on all the days were collected.

Statistical Analysis

Data were analyzed with SPSS version 26.0 (IBM Corp, Armonk, NY). Descriptive statistics, including frequencies of and percentages for categorical variables, mean (±standard deviation; SD) and median (interquartile range; IQR) for continuous variables, were computed for baseline characteristics and PAP parameters. PAP adherence was compared prior to and during lockdown as well as summer and return to school periods using the Friedman test. Mixed-design repeated measures ANOVA subgroup analyses were also performed to compare PAP adherence data across time periods with between-subject factors including sex, diagnoses, e.g obesity and/or underlying medical complexity such as Down syndrome, age, and whether the child was an independent PAP user. Bonferroni post-hoc testing method was performed for statistically significant differences to adjust for multiple comparisons. The degree of change in PAP adherence data between time periods were also compared between different subgroups using the Mann–Whitney *U*-test. Multiple linear regressions to evaluate the association between the percentage of days with a ≥ 4-hour PAP usage and average nightly PAP usage as the primary outcomes, and baseline characteristics as well as days of PAP use.

Ethics

The present study was approved by the Research Ethics Board at The Hospital for Sick Children (REB #1000071460), Toronto, Canada.

Results

Of the 151 in the original study,[23] we were able to obtain data on 149/151 as 2 children had no follow-up PAP usage data available during summer and return to school. In this cohort, 92/149 (61.7%) were male (Table 1). The mean (±SD) age and BMI z-score of the study population were 12.8±4.1 years and 1.45±1.43, respectively. The mean (±SD) duration of prescribed PAP therapy was 3.5 ± 2.7 years. With regards to underlying diagnosis, 53/149 children (35.6%) were obese with no other co-existing medical conditions; 94/149 (63.1%) had an at least one underlying medical condition (eg, Trisomy 21, craniofacial abnormalities, Chiari malformation, achondroplasia, etc.) and the remaining 2/149 (1.3%) were otherwise healthy with no underlying medical diagnosis. In the cohort, 68.5% were prescribed CPAP and 31.5% were prescribed Bi-level PAP. Overall, 35% were described as independent users of PAP and 58.4% had a previous AT.

PAP download data were collected over a mean (±SD) period of 87±11 days prior to lockdown, 89±5 days during lockdown, 89±5 days during summer, and 89±7 days during return to school, respectively (Table 2). Significant differences in PAP usage parameters were found across time periods ($p<0.001$ for all). Specifically, in the cohort, there was a significant increase in median (IQR) percentage of PAP usage for a minimum of 4 hours and average nightly usage during lockdown compared to prior to lockdown. However, during summer and return to school, a further decline was observed across all PAP usage outcome measures, where PAP usage was lower compared to prior to lockdown (Table 2 and Figure 1).

Of the 149 children, 95/149 (64%) showed an increase in PAP usage and the remaining 54/149 (36%) showed a decrease or no change in PAP usage during the first three months of lockdown. Further, 10 (7%) had an abrupt termination in PAP usage during the first three months of lockdown, where 7 (70%) had at least one underlying medical

Table 1 Baseline Demographic and Clinical Characteristics of the Study Population

Characteristics		Participants (N=149)
Age (years)		12.8 ± 4.1
Male, n (%)		92 (61.7)
Weight (kg)		69.6 ± 43.8
Height (cm)		145.7 ± 25.8
Body mass index z-score		1.45 ± 1.43
Duration of non-invasive ventilation (years)		3.5 ± 2.7
Previous adenotonsillectomy, n (%)		87 (58.4)
Independent PAP users, n (%)		52 (34.9)
Race	Caucasian/White, n (%)	55 (36.9)
	Asian, n (%)	32 (21.5)
	African-American/Black, n (%)	23 (15.4)
	Others (eg, Mixed, Native, Hispanic), n (%)	22 (14.8)
	Did not self-identify, n (%)	17 (114)
Diagnosis	Underlying medical conditions, n (%)	94 (63.1)
	Obesity, n (%)	53 (35.6)
	Healthy, n (%)	2 (1.3)
Type of Sleep Disordered Breathing	OSA only, n (%)	109 (73.2)
	OSA and/or CSA with nocturnal hypoventilation	33 (22.1)
	Nocturnal hypoventilation only, n (%)	4 (2.7)
	CSA only, n (%)	3 (2.0)
Type of Non-Invasive Ventilation	CPAP, n (%)	102 (68.5)
	Bi-level PAP, n (%)	47 (31.5)

Note: Data presented as mean ± SD unless otherwise indicated.
Abbreviations: Bi-level PAP, Bi-level positive airway pressure; CPAP, continuous positive airway pressure; CSA, central sleep apnea; OSA, obstructive sleep apnea; PAP, positive airway pressure.

condition and 3 (30%) were obese with no other co-existing medical conditions. PAP usage was resumed among 7/10 (70%), with 3 children restarting during summer and another 4 children restarting after returning to school. Particularly during summer, 76/149 (51%) showed an increase in PAP usage while 73/149 (49%) showed a decrease or no change in PAP usage. An additional 15 children (10%) also abruptly terminated PAP usage. PAP usage was resumed among 6/15 (40%) during return to school. Similarly, abrupt termination of PAP usage was observed among an additional 8 children (5%) during return to school (Figure 2). Interestingly, 3/149 (2%) had used PAP prior to lockdown but stopped using PAP during lockdown and did not restart PAP throughout the rest of the study period.

Overall, during return to school, 69/149 (46%) showed at least sustained PAP usage with the majority, 67/69 (97%) showing increased PAP usage as compared to prior to lockdown. Specifically, those who showed an increase in PAP usage after returning to school showed a median (IQR) increase in average nightly usage of 70 (25–129.5) minutes or a percent change of 22 (10–53)% from prior to lockdown. The remaining 80/149 (54%) had decreased PAP usage during return to school as compared to prior to lockdown, where these children showed a median (IQR) decrease in average

Table 2 Positive Airway Pressure Data Across Time

Measure	Prior to Lockdown (Dec 2019 to March 2020)	During Lockdown (March 2020 to June 2020)	Summer (June 2020 to Sept 2020)	Return to School (Sept 2020 to Dec 2020)	p-value
Number of days of PAP download data collection	87±11 days	89±5 days	89±5 days	89±7 days	0.08
Percentage of days of PAP use (%)	76.0 (36.0–93.7)	87.0 (39.0–100.0)[b, c]	65.5 (10.6–96.3)[b]	72.0 (13.3–98.4)[c]	<0.001[a]
Percentage of days with ≥ 4 hr PAP usage (%)	62.0 (15.8–89.0)[d, e]	77.0 (20.0–94.0)[b, c, d]	44.0 (3.0–85.0)[b, e]	44.0 (6.4–92.5)[c]	<0.001[a]
Average nightly usage (minutes)	367.0 (219.0–500.5)[d]	409.0 (247.5–527.5)[b, c, d]	322.0 (124.0–493.0)[b]	331.0 (100.5–521.0)[c]	<0.001[a]
Average total usage (minutes)	279.0 (99.0–409.0)	310.0 (108.5–443.5)[b]	210.0 (19.5–420.0)[b]	216.0 (39.5–432.5)	<0.001[a]

Notes: Number of days of PAP download data collection presented as mean (± SD). Data presented for N=149 as median (25th-75th percentile) unless otherwise indicated. P-values were calculated using the Friedman test or repeated measures ANOVA based on data distribution, and Bonferroni post-hoc testing was performed for statistically significant differences. [a] p≤0.05 represents a statistically significant difference across time periods. [b] Statistically significant difference between during lockdown and summer periods. [c] Statistically significant difference between during lockdown and return to school periods. [d] Statistically significant difference between prior to lockdown and during lockdown periods. [e] Statistically significant difference between prior to lockdown and summer periods. Prior to lockdown: December 18, 2019-March 16, 2020. During lockdown: March 17-June 14, 2020. Summer: June 15-September 13, 2020. Return to school: September 14-December 12, 2020. Percentage of days of PAP use – Overall percentage of days that PAP was used regardless of duration of usage. Percentage of days with ≥ 4 hr PAP usage – Percentage of days where PAP was used for ≥ 4 hours usage. Average nightly usage – Average minutes of PAP use based on days where PAP was used. Average total usage – Average minutes of PAP use based on all the days over which the data was collected.

nightly usage of −91 (−171 - −32.3) minutes or percent change of −30 (−99 - −11)% from prior to lockdown. Of the 80 children who showed decreased PAP usage, 20/80 (25%) or 13.4% of the entire cohort, showed termination of PAP. Among these 20 children, 12/20 (60%) were obese with no other co-existing medical conditions.

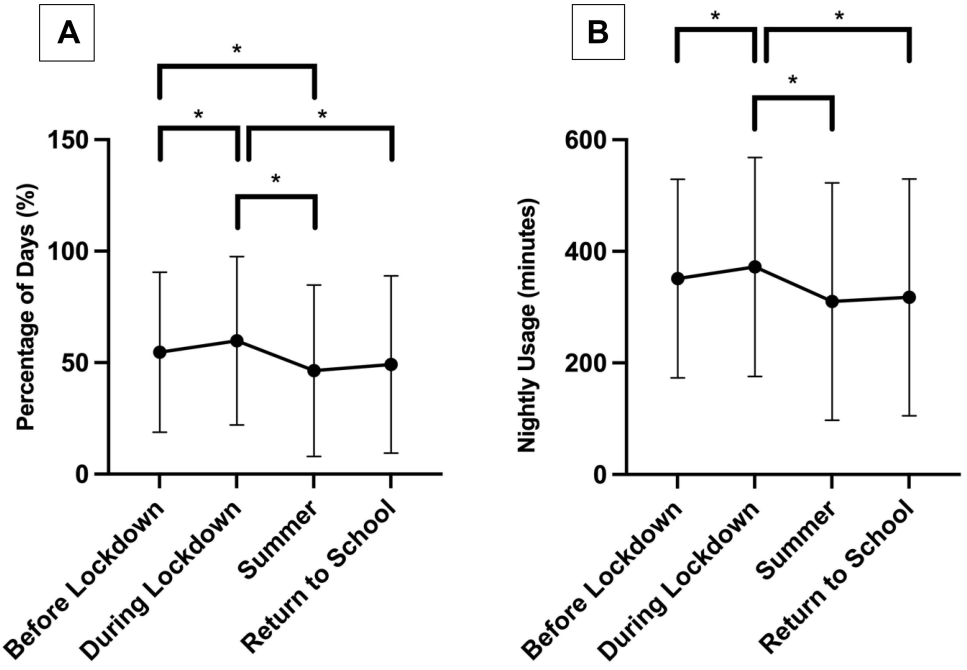

Figure 1 Primary outcomes for PAP adherence use across time periods. Median and IQR values are shown for (**A**) percentage of days with a minimum of 4-hour PAP usage, and (**B**) average nightly usage of PAP (eg, average minutes of PAP use based on days where PAP was used), across four time periods. *Indicates significant differences between the corresponding time periods (p<0.05).

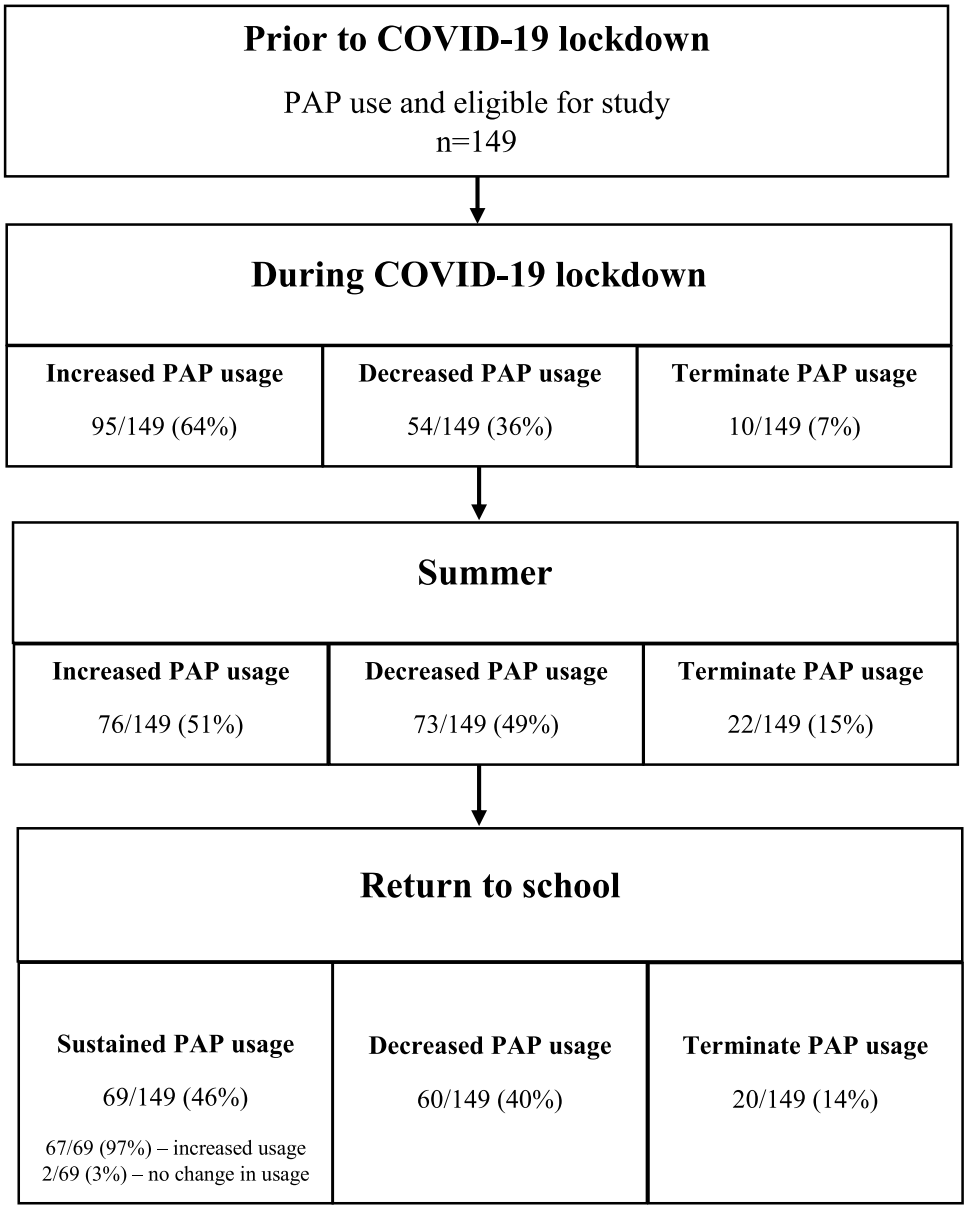

Figure 2 Flow chart of subject recruitment and changes in PAP usage across the 9-month period following the start of the COVID-19 lockdown.

Further subgroup analyses were also performed, where the primary outcomes of PAP adherence were compared between and within different subgroups including 1) underlying diagnoses, children with obesity versus children with underlying medical complexity (with and without obesity) 2) males versus females, 3) children who were 12 years and younger, between 12 and 15 years old, or 15 years and older, and 4) CPAP or Bi-level PAP users (Supplementary e-Tables 1–4, respectively, and Figure 3). After adjusting for use of PAP prior to the lockdown, percentage of days with a minimum of 4-hour PAP usage was significantly higher for those with underlying medical conditions compared to children with obesity prior to lockdown (p=0.009), during lockdown (p=0.001), summer (p=0.02) and return to school (p=0.007) (Supplementary e-Table 1 and Figure 3A). However, the absolute degree of change (hours) in the minimum of 4-hour PAP usage did not significantly differ between both groups across time (during lockdown: p=0.29; summer: p=0.85; return to school: p=0.18). Similarly, no significant differences were found with percent change in the minimum of 4-hour PAP usage between those with underlying medical conditions and those with obesity (during lockdown:

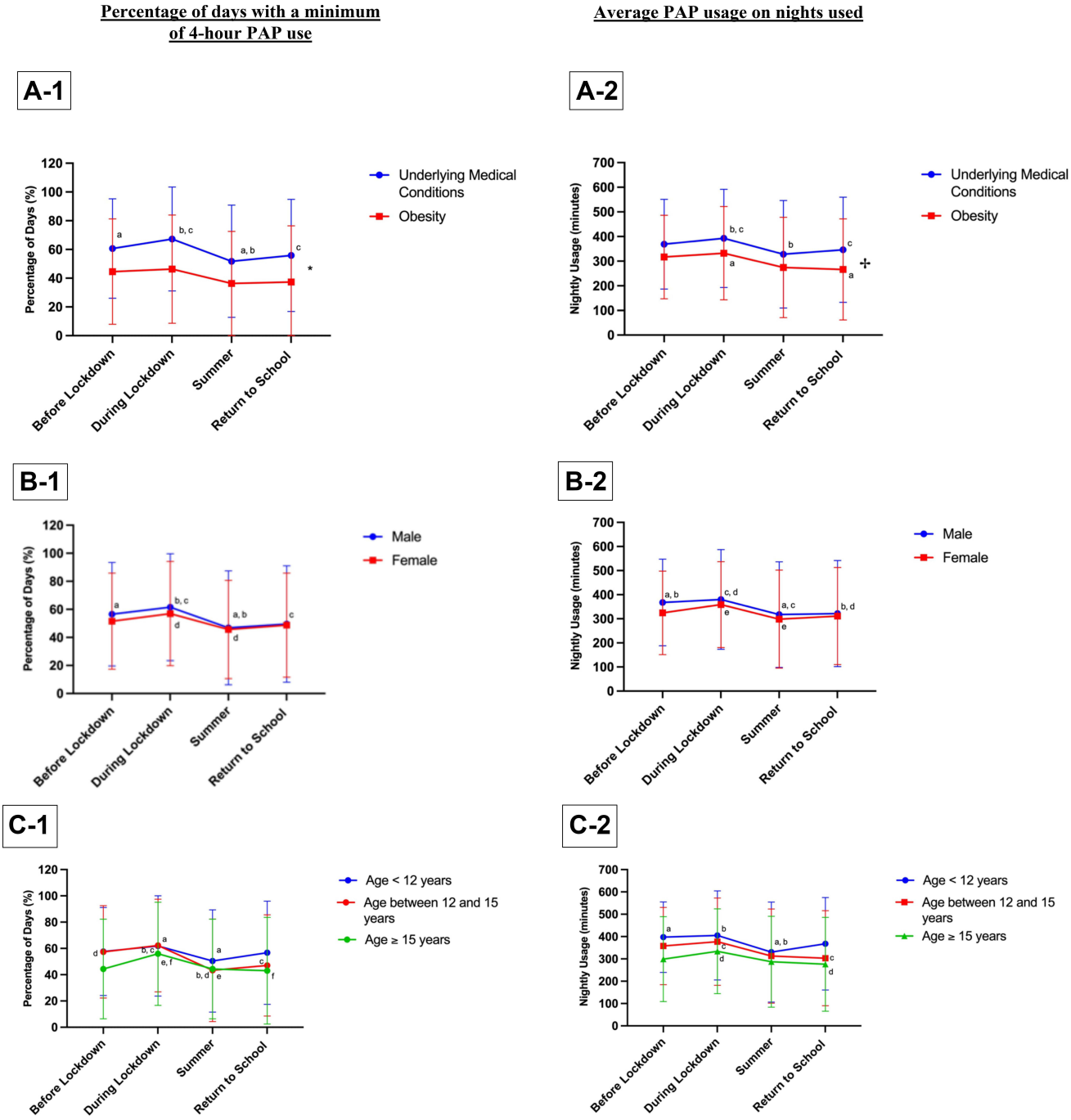

Figure 3 Continued.

p=0.78; summer: p=0.60; return to school: p=0.15). No significant between-group differences were found for either primary outcome between males and females, age groups as well as CPAP and Bi-level PAP users (Figure 3B–D).

PAP usage was also evaluated among independent and dependent PAP users, with dependent PAP users showing significantly higher percentage of days with a minimum of 4-hour PAP usage prior to lockdown (p=0.007), during lockdown (p=0.02), summer (p=0.01) and return to school (p<0.001) (Figure 3E). There were no significant differences between independent or dependent PAP users across time in either the absolute degree of change (hours) (during lockdown: p=0.75; summer: p=0.76; return to school: p=0.07) or the percent change in the minimum of 4-hour PAP usage (during lockdown: p=0.87; summer: p=0.45; return to school: p=0.15). Similarly, dependent PAP users

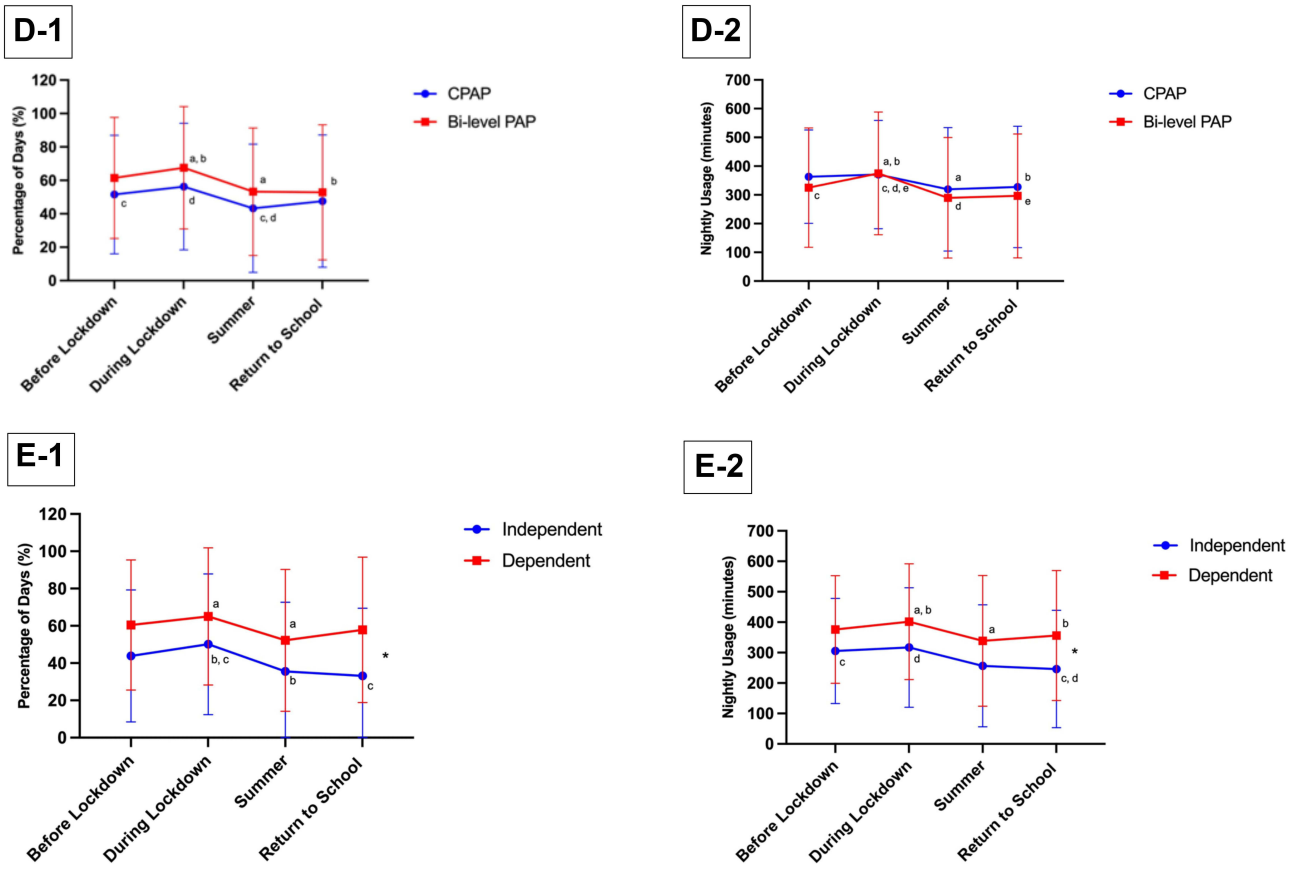

Figure 3 Percentage of days with a minimum of 4-hour PAP use and average PAP usage on nights used (eg, average minutes of PAP use based on days where PAP was used) across time periods for different subgroups. Mean and SD values for (**A**) children with underlying medical conditions versus obesity, (**B**) males versus females, (**C**) those aged <12 years, between 12 and 15 years, and ≥15 years, (**D**) those who used CPAP versus Bi-level PAP, and (**E**) independent versus dependent users, across four time periods (before lockdown, during the first 3 months of lockdown, summer time and return to school). Percentage of days with a minimum of 4-hour use and average PAP usage on nights used are shown on the left and the right, respectively. Similar letters denote significant differences between the corresponding time periods within each group. The following symbols indicate significant differences between groups at the following periods: * - each of the four time periods, and ✢ - return to school.

demonstrated higher average nightly usage prior to lockdown (p=0.02), during lockdown (p=0.01), summer (p=0.02) and return to school (p=0.002). However, no significant differences were found between groups across time in the absolute degree of change (minutes) (during lockdown: p=0.72; summer: p=0.46; return to school: p=0.11) or the percent change in average nightly usage (during lockdown: p=0.92; summer: p=0.41; return to school: p=0.09). Within the dependent group, PAP usage was compared between children with and without development delay (Supplementary e-Table 5). There were no significant between-group differences; however those with developmental delay showed significantly higher percentage of days with a minimum of 4-hour PAP usage (p=0.02) and average nightly usage (p=0.05) during lockdown compared to the summer period.

In terms of within-group differences, a similar trend was observed among most subgroups where the percentage of days with a minimum of 4-hour PAP usage and average nightly usage was significantly higher during lockdown (p<0.05) compared to either prior to lockdown, summer or return to school. In children with obesity, average nightly usage was significantly higher during lockdown compared to return to school (p=0.02); however, no significant differences were found in the percentage of days with a minimum of 4-hour PAP usage in this subgroup across the time periods (Supplementary e-Table 1 and Figure 3A).

Previously, using hierarchical multiple regression models, we found that children with obesity were more likely to show a decline in the percentage of days with a minimum of 4-hour PAP usage during lockdown compared to those with underlying medical conditions. Similar models were performed to determine associations between the percentage of days with 4-hour PAP usage during summer and return to school as the dependent variables. Baseline characteristics that can

affect PAP use such as age (years), diagnosis (obese or other medical conditions), sex (male or female) and PAP type (CPAP or Bi-level PAP) as well as the use of PAP prior to the lockdown were included in the regression models. No significant associations were found during summer; however, during return to school, a significant association was found where children with obesity were more likely to demonstrate a decrease in the percentage of days with 4-hour PAP usage compared to those with underlying medical conditions (β=−15.36, p=0.03) (Table 3). Using similar models with average nightly PAP usage as the dependent variable, no significant associations were found during summer or return to school.

Discussion

To our knowledge, there are no current published studies that have evaluated the longitudinal trajectory of PAP usage in children since the pandemic has begun. Our principal finding demonstrated that 1) PAP adherence in children with SDB decreased across 9 months period after the start of COVID-19 lockdown, where 54% showed an overall decrease in PAP usage by the end of the 9-month period; 2) Dependent PAP users had significantly higher PAP usage compared to independent PAP users; 3) Children with underlying medical conditions had greater number of days of PAP usage; and 4) Obesity was a risk factor for a decline in PAP usage. Risk factors associated with decline in PAP use are well documented which includes independent status and lack of support from family or health-care team,[11,15,28,29] discomfort of the PAP device (mask leak, PAP pressure and tubing),[8,16,29] negative experiences or attitude with PAP,[28] difficulty adapting,[28] and a lack of subjective symptom improvement[16,28,29] as well as fear or embarrassment regarding treatment.[28] Our study showed a significant further decline from baseline PAP usage during specific time periods of the COVID-19 pandemic which suggest that there are additional risk factors during this time periods.

Despite the fact that PAP is an effective treatment to minimize symptoms of SDB, PAP therapy is not always well tolerated in both pediatric and adult populations and long-term adherence to treatment can be problematic.[11–14,30] A comprehensive systematic literature review of 82 adult studies regarding CPAP adherence over a twenty year time-frame (1994–2015) found that the rate of CPAP adherence remained persistently low over twenty years with PAP non-adherence rate of 34.1%.[30] Similarly, in children, PAP adherence rates as determined by objective analyses over 20,000 children prior to the pandemic was 61.8%[8] which is similar to our data at the start of the pandemic.

However, of concern, our data showed that there was a further decline in PAP usage during the COVID-19 pandemic. The reasons for changes in PAP adherence over time in our cohort are likely to be multifactorial including attendance of in-person versus online school as well as COVID-19 pandemic and restrictive measurements.[24,25,31–34] These changes have impacted sleep routine schedules while also resulting in poorer quality of sleep, decreased physical activity, increase in sedentary behavior and screen time as well as adverse psychological and mental health effects on children and caregivers.[24,25,31] A recent study examined the impact of the COVID-19 pandemic on the 24 hours movement behaviors of 2661 Canadian students in grades 7–11 (43.4% male) over a one year period in Montral, Quebec, Canada.[32] The study demonstrated that the variability in the 24 hour movement behaviors were consistent with the restrictive measures imposed by the government as well as the COVID-19 pandemic direct and indirect effects.[32] It was observed that most

Table 3 Multiple Regression Analysis Assessing Factors Associated with Percentage of Days with a Minimum 4-Hour Usage of Positive Airway Pressure After Returning to School

Parameters	Coefficients	Standard Error	*p*-value
Age	−1.23	0.81	0.13
Diagnosis (eg, Obesity or Underlying Medical Conditions)	−15.36	7.12	0.03[a]
Sex (Male or Female)	−1.75	6.74	0.80
PAP Type (eg, CPAP or Bi-level PAP)	−1.11	7.10	0.88
Number of days of PAP download data collection prior to lockdown	0.45	0.30	0.13

Notes: P-values were calculated using linear regression. [a]p≤0.05 represents statistical significance. Model shows significant association with obesity, suggesting that children with obesity were more likely to demonstrate a decrease in the percentage of days with 4-hour PAP usage compared to those with underlying medical conditions (p=0.03).
Abbreviations: Bi-level PAP, Bi-level positive airway pressure; CPAP, continuous positive airway pressure; PAP, positive airway pressure.

children and adolescents had highest percentage of increased recreational screen time (68%) with the highest percentage (55%) of decreased physical activity between March 13 to June 2020, which was consistent with the timing of the COVID-19 lockdown that started on March 13, 2020 that led to severe restrictive measures and high schools closures with remote learning. Moreover, 49% of the participants reported a decrease in their sleep duration and 47% of them experienced poor sleep quality during this time,[32] factors which may have impacted PAP use in our cohort. Other factors that may have influenced PAP adherence may include seasonal variation as adolescents have earlier bedtimes in the winter and spring compared to later bedtimes in the summer.[34–36] A recent study among adults with OSA also demonstrated that seasonal changes were associated with CPAP adherence.[37] In this study, adults who were adherent to CPAP (eg, 70% of PAP use for ≥ 4 hours over a one-year period) showed both a lower percentage of CPAP use for ≥ 4 hours daily and daily duration of CPAP use during the summer compared to other seasons. Among adults who were non-adherent to CPAP, the percentage of CPAP use for ≥ 4 hours daily was greater in the spring compared to other seasons, with the daily duration of CPAP use being the shortest in the summer and longest in the winter.[37] Our results showed a similar trend with PAP usage during the pandemic, where children showed lower PAP usage in the summer compared to the spring and winter months.

Despite overall decreases in PAP adherence over time, dependent PAP users had significantly higher usage of PAP compared to independent PAP users which concurs with previously reported data in 240 children (median age 7.9 years) of which 103 (42.9%) children had developmental disabilities.[38] Furthermore, a retrospective study of CPAP adherence in 140 children (aged 12.0 ± 5.7 years old) over a one-year period showed CPAP adherence was higher in those with developmental delay, possibly due to their physical or intellectual disability resulting in a reduced ability to remove the CPAP mask and increased caregiver support. Moreover, during the pandemic, parental perception and beliefs of PAP necessity for those with neurodevelopmental disabilities may have increased.[11]

We found that obesity was a risk factor for decreased PAP usage over time, similar to a previous 10-year multicentred retrospective review that evaluated longitudinal PAP adherence among 622 children aged 0–18 years old, which showed that children with obesity or metabolic syndrome were more likely to decline PAP therapy (OR =2.077 [95% CI 1.110–3.888]).[39] The reasons for these findings are unclear; however, there is one study suggesting lack of perceived benefits of PAP in this population may contribute to decreased use.[16] Of concern, obesity is an independent risk factor for susceptibility to and severity of COVID-19 which is related to worsening outcomes including increased risk of hospitalization, critical care admission and mortality.[40,41] The potential mechanisms include chronic inflammation, impairment of respiratory function and pulmonary perfusion, impaired immune response and underlying cardiorespiratory diseases.[40,41] Additionally, OSA has been reported as a risk factor for severe COVID-19.[42] A study of 445 adults with COVID-19 (aged 55.1±8.0 years old, 81.8% male and 8.5% diagnosed OSA) demonstrated that OSA is an independent risk factor for severe COVID- 19 and associated with higher risk for COVID-19 hospitalization (OR 2.93, 95% CI 1.02 to 8.39, p=0.045).[42] Therefore, children with obesity, diagnosed SDB and poor PAP adherence are likely at greater risk for worsening COVID-19 outcomes and heightened awareness among physicians is paramount for early education, counselling and necessary interventions to encourage long-term PAP adherence.

Other risk factors for consideration includes both access and delivery of health care which has significantly pivoted and changed worldwide during the current COVID-19 pandemic. From mid-March 2020, there has been an observed 80% decrease in office visits and 56-fold increase in virtual care visits.[18] The benefits of telemedicine including telediagnosis, teletherapy, telemonitoring and teleconsultation have reduced waiting times, transportation time, visit costs and lowered infections rates,[43–48] which may also better accomodate schedules of families while reducing the need to take time off work or arranging for childcare. However, the disadvantages of telemedicine includes the inability to perform most physical examinations or procedures, difficulty establishing new therapeutic relationships which are especially important for sustaining PAP use and reinforcing education around sleep hygiene issues particularly during adolescence, dealing with certain complex mental health issues, difficulty with interpreting body language and nonverbal clues, and lacking the full degree of comfort and support that can be provided in person as well as privacy and continuity of care concerns.[18] For children with SDB, in-person visits are vital to troubleshoot PAP related issues such as adequate mask fittings, equipment issues and additional face-to-face education with both caregivers and children themselves.[44] Importantly, direct interactions can emphasize the critical role of the caregiver especially in the adolescent population.[43]

The strengths of this study include a large time frame for longitudinal follow-up during consecutive 3-month periods to compare changes in PAP adherence through the COVID-19 pandemic. Further, the heterogeneity of participants suggests that these results can be generalizable across paediatric cohorts of children with SDB using PAP therapy. Our results were also presented as group level means, which may result in individual changes to be missed; however by performing repeated measures ANOVA analyses, such limitations of group level means were minimized as our findings considered both within-subject variation and between-group effects. Additionally, we were able to obtain objective data from PAP devices which have been validated to be accurate. Our study highlights that longitudinal PAP adherence requires a long-term approach with frequent education and counselling, clinical monitoring, treatment strategies or other targeted behavioral interventions,[29] parental engagement,[16,29] social support[29] and peer-driven intervention[49] to prevent further decline in PAP use of children with SDB.

There are some notable limitations to our study which require consideration. Factors influencing PAP adherence across all four distinct time periods were not available. These factors include bed time, wake time, sleep duration and sleep disturbance, as well as psychosocial factors, mental health, family functioning, change in caregiver schedules, all of which can influence PAP usage. Moreover, given the retrospective nature of our study, we do not have information on the COVID-19 positivity status of our participants. During clinic visits, we were able to ask 100/149 participants whether they had tested positive for COVID-19 and we know that 2/100 did state they had been positive, where one child continued to use PAP throughout all four time periods, and the second child stopped using PAP upon returning to school. However, this information was not confirmed by reviewing results of PCR testing and we also do not know the timing of the COVID-19 positive testing in relation to obtaining the PAP data. Moreover, we do not know whether any family members tested positive for COVID-19. Our study is also limited as we did not collect and therefore were unable to compare the PAP usage across a one-year period prior to COVID-19 pandemic. Further, we were not able to identify additional factors as to why children stopped using PAP as we do not have detailed survey or qualitative data available. However, these data highlight the importance of future research to identify and mitigate the factors associated with decreased PAP usage in children with SDB.

Conclusion

Untreated or undertreated SDB is associated with significant adverse physical, physiological and psychosocial outcomes. PAP adherence has been significantly impacted by the COVID-19 pandemic. Overall, there was decline in PAP adherence over time during the COVID-19 pandemic when compared to pre-pandemic levels, leaving many children untreated for SDB. Our study emphasized the need for physicians to detail the barriers and facilitators in order to provide targeted therapeutic interventions for reinforcing long-term PAP adherence in children with SDB. Future multicentered studies should address whether behavioral and/or additional virtual care modifications can be utilized and optimized to improve long-term PAP adherence and clinical outcomes in children with SDB during both the pandemic and beyond.

Abbreviations

AHI, apnea-hypopnea index; AT, adenotonsillectomy; Bi-level PAP, Bi-level positive airway pressure; BMI, body mass index; CPAP, continuous positive airway pressure; CSA, central sleep apnea; IQR, interquartile range; NH, nocturnal hypoventilation; OSA, obstructive sleep apnea; PAP, positive airway pressure; PSG, polysomnography; SARS-CoV-2, severe acute respiratory syndrome coronavirus 2; SDB, sleep disordered breathing; SD, standard deviation.

Data Sharing Statement

All data relevant to the study are included in the article or available in the Supplementary materials.

Ethics Approval

This study was approved by the SickKids Research Ethics Board (REB No. 1000071460). Parental and/or participant consent for reviewing patient medical records was not required by the SickKids Research Ethics Board due to retrospective study. Patient data confidentiality and compliance were covered by the Declaration of Helsinki.

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Disclosure

The authors have no conflicts of interest for this work to declare.

References

1. Castro-Codesal ML, Dehaan K, Featherstone R, et al. Long-term non-invasive ventilation therapies in children: a scoping review. *Sleep Med Rev*. 2018;37:148–158. doi:10.1016/j.smrv.2017.02.005
2. Fedor KL. Noninvasive respiratory support in infants and children. *Respir Care*. 2017;62(6):699–717. doi:10.4187/respcare.05244
3. Amin R, Al-Saleh S, Narang I. Domiciliary noninvasive positive airway pressure therapy in children. *Pediatr Pulmonol*. 2016;51(4):335–348. doi:10.1002/ppul.23353
4. Parmar A, Baker A, Narang I. Positive airway pressure in pediatric obstructive sleep apnea. *Paediatr Respir Rev*. 2019;31:43–51. doi:10.1016/j.prrv.2019.04.006
5. Marcus CL, Radcliffe J, Konstantinopoulou S, et al. Effects of positive airway pressure therapy on neurobehavioral outcomes in children with obstructive sleep apnea. *Am J Respir Crit Care Med*. 2012;185(9):998–1003. doi:10.1164/rccm.201112-2167OC
6. DelRosso LM, King J, Ferri R. Systolic blood pressure elevation in children with obstructive sleep apnea is improved with positive airway pressure use. *J Pediatr*. 2018;195(102):102–107.e1. doi:10.1016/j.jpeds.2017.11.043
7. Ramirez A, Khirani S, Aloui S, et al. Continuous positive airway pressure and noninvasive ventilation adherence in children. *Sleep Med*. 2013;14(12):1290–1294. doi:10.1016/j.sleep.2013.06.020
8. Bhattacharjee R, Benjafield AV, Armitstead J, et al. Adherence in children using positive airway pressure therapy: a big-data analysis. *Lancet Digit Health*. 2020;2(2):e94–e101. doi:10.1016/S2589-7500(19)30214-6
9. Marcus CL, Rosen G, Ward SL, et al. Adherence to and effectiveness of positive airway pressure therapy in children with obstructive sleep apnea. *Pediatrics*. 2006;117(3):e442–51. doi:10.1542/peds.2005-1634
10. Simon SL, Duncan CL, Janicke DM, Wagner MH. 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):172–177. doi:10.1016/j.sleep.2011.10.026
11. Hawkins SM, Jensen EL, Simon SL, Friedman NR. Correlates of pediatric CPAP adherence. *J Clin Sleep Med*. 2016;12(6):879–884. doi:10.5664/jcsm.5892
12. Blinder H, Momoli F, Holland SH, Blinder A, Radhakrishnan D, Katz SL. Clinical predictors of nonadherence to positive airway pressure therapy in children: a retrospective cohort study. *J Clin Sleep Med*. 2021;17(6):1183–1192. doi:10.5664/jcsm.9162
13. Marcus CL, Beck SE, Traylor J, et al. Randomized, double-blind clinical trial of two different modes of positive airway pressure therapy on adherence and efficacy in children. *J Clin Sleep Med*. 2012;8(1):37–42. doi:10.5664/jcsm.1656
14. Puri P, Ross KR, Mehra R, et al. Pediatric positive airway pressure adherence in obstructive sleep apnea enhanced by family member positive airway pressure usage. *J Clin Sleep Med*. 2016;12(7):959–963. doi:10.5664/jcsm.5924
15. Parmar A, Messiha S, Baker A, Zweerink A, Toulany A, Narang I. Caregiver support and positive airway pressure therapy adherence among adolescents with obstructive sleep apnea. *Paediatr Child Health*. 2020;25(8):491–497. doi:10.1093/pch/pxz107
16. Alebraheem Z, Toulany A, Baker A, Christian J, Narang I. Facilitators and barriers to positive airway pressure adherence for adolescents. A qualitative study. *Ann Am Thorac Soc*. 2018;15(1):83–88. doi:10.1513/AnnalsATS.201706-472OC
17. Pollard CA, Morran MP, Nestor-Kalinoski AL. The COVID-19 pandemic: a global health crisis. *Physiol Genomics*. 2020;52(11):549–557. doi:10.1152/physiolgenomics.00089.2020
18. Glazier RH, Green ME, Wu FC, Frymire E, Kopp A, Kiran T. Shifts in office and virtual primary care during the early COVID-19 pandemic in Ontario, Canada. *Can Med Assoc J*. 2021;193(6):E200–e210. doi:10.1503/cmaj.202303
19. Miller MA, Cappuccio FP. A systematic review of COVID-19 and obstructive sleep apnoea. *Sleep Med Rev*. 2020;55:101382. doi:10.1016/j.smrv.2020.101382
20. Johnson KG, Sullivan SS, Nti A, Rastegar V, Gurubhagavatula I. The impact of the COVID-19 pandemic on sleep medicine practices. *J Clin Sleep Med*. 2021;17(1):79–87. doi:10.5664/jcsm.8830
21. Attias D, Pepin JL, Pathak A. Impact of COVID-19 lockdown on adherence to continuous positive airway pressure by obstructive sleep apnoea patients. *Eur Respir J*. 2020;56(1):2001607. doi:10.1183/13993003.01607-2020
22. Demirovic S, Lusic Kalcina L, Pavlinac Dodig I, et al. The COVID-19 lockdown and CPAP adherence: the more vulnerable ones less likely to improve adherence? *Nat Sci Sleep*. 2021;13:1097–1108. doi:10.2147/nss.s310257
23. Sunkonkit K, Selvadurai S, Voutsas G, et al. The impact of the COVID-19 pandemic on positive airway pressure usage in children with sleep-disordered breathing. *Sleep Breath*. 2021. doi:10.1007/s11325-021-02409-w
24. Bates LC, Zieff G, Stanford K, et al. COVID-19 impact on behaviors across the 24-hour day in children and adolescents: physical activity, sedentary behavior, and sleep. *Children*. 2020;7(9):Sep. doi:10.3390/children7090138
25. Dhiman S, Sahu PK, Reed WR, Ganesh GS, Goyal RK, Jain S. Impact of COVID-19 outbreak on mental health and perceived strain among caregivers tending children with special needs. *Res Dev Disabil*. 2020;107:103790. doi:10.1016/j.ridd.2020.103790

26. Centers for Disease Control and Prevention. Defining childhood obesity. Available from: https://www.cdc.gov/obesity/childhood/defining.html. Accessed April 2, 2022.
27. ResMed. AirView. Available from: https://www.resmed.com/epn/en/healthcare-professional/products/monitoring-and-data-management/airview-patient-management-system.html. Accessed November 22, 2020.
28. Ennis J, Rohde K, Chaput JP, Buchholz A, Katz SL. Facilitators and barriers to noninvasive ventilation adherence in youth with nocturnal hypoventilation secondary to obesity or neuromuscular disease. *J Clin Sleep Med*. 2015;11(12):1409–1416. doi:10.5664/jcsm.5276
29. Sawyer AM, Gooneratne NS, Marcus CL, Ofer D, Richards KC, Weaver TE. A systematic review of CPAP adherence across age groups: clinical and empiric insights for developing CPAP adherence interventions. *Sleep Med Rev*. 2011;15(6):343–356. doi:10.1016/j.smrv.2011.01.003
30. Rotenberg BW, Murariu D, Pang KP. Trends in CPAP adherence over twenty years of data collection: a flattened curve. *J Otolaryngol Head Neck Surg*. 2016;45(1):43. doi:10.1186/s40463-016-0156-0
31. Moore SA, Faulkner G, Rhodes RE, et al. Impact of the COVID-19 virus outbreak on movement and play behaviours of Canadian children and youth: a national survey. *Int J Behav Nutr Phys Act*. 2020;17(1):85. doi:10.1186/s12966-020-00987-8
32. Dubuc MM, Berrigan F, Goudreault M, Beaudoin S, Turcotte S. COVID-19 impact on adolescent 24 h movement behaviors. *Int J Environ Res Public Health*. 2021;18(17):9256. doi:10.3390/ijerph18179256
33. Pietrobelli A, Pecoraro L, Ferruzzi A, et al. Effects of COVID-19 lockdown on lifestyle behaviors in children with obesity living in Verona, Italy: a longitudinal study. *Obesity*. 2020;28(8):1382–1385. doi:10.1002/oby.22861
34. Quante M, Wang R, Weng J, et al. Seasonal and weather variation of sleep and physical activity in 12–14-year-old children. *Behav Sleep Med*. 2019;17(4):398–410. doi:10.1080/15402002.2017.1376206
35. Hjorth MF, Chaput JP, Michaelsen K, Astrup A, Tetens I, Sjödin A. Seasonal variation in objectively measured physical activity, sedentary time, cardio-respiratory fitness and sleep duration among 8–11 year-old Danish children: a repeated-measures study. *BMC Public Health*. 2013;13(1):808. doi:10.1186/1471-2458-13-808
36. Moreno JP, Razjouyan J, Lester H, et al. Later sleep timing predicts accelerated summer weight gain among elementary school children: a prospective observational study. *Int J Behav Nutr Phys Act*. 2021;18(1):94. doi:10.1186/s12966-021-01165-0
37. Fujino Y, Oka Y, Wakamura T. Seasonal effects on the continuous positive airway pressure adherence of patients with obstructive sleep apnea. *Sleep Med*. 2021;80:126–133. doi:10.1016/j.sleep.2021.01.024
38. Kang EK, Xanthopoulos MS, Kim JY, et al. 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. doi:10.5664/jcsm.7850
39. Bedi PK, DeHaan K, MacLean JE, Castro-Codesal ML. Predictors of longitudinal outcomes for children using long-term noninvasive ventilation. *Pediatr Pulmonol*. 2021;56(5):1173–1181. doi:10.1002/ppul.25188
40. Kwok S, Adam S, Ho JH, et al. Obesity: a critical risk factor in the COVID-19 pandemic. *Clin Obes*. 2020;10(6):e12403. doi:10.1111/cob.12403
41. Nogueira-de-almeida CA, Del Ciampo LA, Ferraz IS, Del Ciampo IRL, Contini AA, Ued FDV. COVID-19 and obesity in childhood and adolescence: a clinical review. *J Pediatr (Rio J)*. 2020;96(5):546–558. doi:10.1016/j.jped.2020.07.001
42. Strausz S, Kiiskinen T, Broberg M, et al. Sleep apnoea is a risk factor for severe COVID-19. *BMJ Open Respir Res*. 2021;8(1):e000845. doi:10.1136/bmjresp-2020-000845
43. Voulgaris A, Ferini-Strambi L, Economou NT, Nena E, Steiropoulos P. Sleep telemedicine practices: checkpoint list and practical considerations in the COVID-19 era. *Front Neurol*. 2021;12:664274. doi:10.3389/fneur.2021.664274
44. Paruthi S. Telemedicine in pediatric sleep. *Sleep Med Clin*. 2020;15(3s):e1–e7. doi:10.1016/j.jsmc.2020.07.003
45. Schöbel C, Werther S, Teschler H, Taube C. Telemedicine in respiratory sleep medicine: COVID-19 pandemic unmasks the need for a process-oriented, replicable approach for implementation in clinical routine. *J Thorac Dis*. 2020;12(Suppl 2):S261–s263. doi:10.21037/jtd-cus-2020-011
46. Voulgaris A, Ferini-Strambi L, Steiropoulos P. Sleep medicine and COVID-19. Has a new era begun? *Sleep Med*. 2020;73:170–176. doi:10.1016/j.sleep.2020.07.010
47. Zia S, Fields BG. Sleep telemedicine: an emerging field's latest frontier. *Chest*. 2016;149(6):1556–1565. doi:10.1016/j.chest.2016.02.670
48. Reed ME, Huang J, Graetz I, et al. Patient characteristics associated with choosing a telemedicine visit vs office visit with the same primary care clinicians. *JAMA Netw open*. 2020;3(6):e205873. doi:10.1001/jamanetworkopen.2020.5873
49. Parthasarathy S, Wendel C, Haynes PL, Atwood C, Kuna S. A pilot study of CPAP adherence promotion by peer buddies with sleep apnea. *J Clin Sleep Med*. 2013;9(6):543–550. doi:10.5664/jcsm.2744