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ORIGINAL ARTICLE

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Analysis of mandibular jaw movements to assess ventilatory support management of children with obstructive sleep apnea syndrome treated with positive airway pressure therapies

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Abstract

Background: The polysomnography (PSG) is the gold-standard for obstructive sleep apnea (OSA) syndrome diagnosis and assessment under positive airway pressure (PAP) therapies in children. Recently, an innovative digital medicine solution, including a mandibular jaw movement (MJM) sensor coupled with automated analysis, has been validated as an alternative to PSG for pediatric application.

Objective: This study aimed to assess the reliability of MJM automated analysis for the assessment of residual apnea/hypopnea events during sleep in children with OSA treated with noninvasive ventilation (NIV) or continuous PAP (CPAP).

Methods: In this open-label prospective non-randomized multicentric trial, we included children aged from 5 to 18 years with a diagnosis of severe OSA. The children underwent in-laboratory PSG with simultaneous MJM monitoring and at-home recording with MJM monitoring 3 months later. Agreement between PSG and MJM analysis in measuring the residual apnea-hypopnea index (AHI) was evaluated by the Bland–Altman method. The treatment effect on residual AHI was estimated for both PSG and MJM analysis.

Results: Fifteen (60% males) children were included with a median age of 12 years [interquartile range 8–15]. Two (17%) were ventilated with NIV and 13 (83%) with CPAP. There was a good agreement between MJM-AHI and PSG-AHI with a median bias of −0.25 (95% CI: −3.40 to +2.04) events/h. The reduction in AHI under treatment was consistently significant across the three measurement methods: in-laboratory PSG and MJM recordings in the laboratory and at home.

Conclusion: Automated analysis of MJM is a highly reliable alternative method to assess residual events in a small population treated with PAP therapies.

Guillaume Mortamet and Jean Louis Pépin are cosenior authors.

KEYWORDS
continuous positive airway pressure, machine learning, mandibular jaw movement, noninvasive ventilation, pediatric obstructive sleep apnea

1 | INTRODUCTION

Obstructive sleep apnea (OSA) syndrome is a common condition in children with an estimated prevalence of 1%–4%.[1,2] There is a peak in the frequency between the ages of 3 and 6 years in relation to physiological tonsil hypertrophy.[3] There are also predisposing factors such as obesity, upper airway infections, prematurity, and asthma.[2] In addition, many congenital or acquired pathologies are associated with OSA in the pediatric population, including syndromic, neuromuscular, respiratory, or metabolic pathologies.[4–6]

The polysomnography (PSG) remains the gold-standard for the diagnosis of sleep-disordered breathing in children but is expensive, time consuming, requires expertise for scoring, and is not widely available.[7,8] For these reasons, there is a consensus on the need to develop simpler and less expensive diagnostic approaches to improve access to care.[8] Previous attempts to achieve these goals have included questionnaires or nocturnal pulse oximetry, which have limited success.[7,9,10]

The American Academy of Sleep Medicine (AASM) does not currently endorse the use of home sleep tests for the diagnosis of OSA in children due to insufficient scientific evidence regarding the feasibility and validity of the tests.[11] Recently, an innovative digital medicine solution, including mandibular jaw movement (MJM) monitoring and artificial intelligence, has been developed and validated in adults.[12] Another study using this technology coupled with automated analysis conducted in the pediatric population also demonstrated a good agreement between MJM and the polysomnographic results.[13]

The PSG is also the reference technique for the evaluation of treatment efficacy and residual events in children treated by continuous positive airway pressure (CPAP) or noninvasive ventilation (NIV). The management of OSA in children is essentially based on the treatment of the underlying cause of obstruction (tonsillectomy or maxilla facial surgery, orthodontic management, allergy management, weight loss in case of obesity).[14,15] When access or effectiveness are limited or in case of moderate to severe OSA, CPAP, or NIV are indicated. However, such therapies can be poorly tolerated and suboptimal, leading to ventilation failure.[16]

Beyond PSG, there are limited techniques to assess CPAP or NIV efficacy, although output data can be used to monitor treatment in routine practice.[16–19] Therefore, MJM analysis could be adopted as an additional alternative approach to monitor treatment outcomes in children.

In the present study, we aimed to assess the reliability of measurements of MJM supported by automated analysis for identifying residual apnea-hypopnea index (AHI) in children with OSA treated by CPAP or NIV.

2 | METHODS

2.1 | Design

This prospective, multicentric, open-label study was conducted at the University Hospital of Grenoble Alpes (center 1) and the University Armand Trousseau Hospital, AP-HP, in Paris (center 2) from March 2022 to June 2023. Data were collected during two nights: at the hospital (concurrent recording of PSG and MJM) and at home 3 months later (only MJM). Written informed consents were obtained from all children and legal-representatives. The protocol for this study was approved by the Ile de France IV Institutional Review Board and the Medical Ethics Committee (ID-RCB: 2021-A02697-34).

2.2 | Participants

Participants were aged 5–18 years with severe OSA at baseline and treated by NIV or CPAP during sleep with a nasal or nostril interface. OSA type 1 corresponded to tonsil hypertrophy, type 2 to patients with obesity, and type 3 to children with polymalformative syndromes. Severe OSA was diagnosed during a previous PSG (defined by an AHI ≥ 10/h or an apnea index (AI) ≥ 5/h), and associated or not with alveolar hypoventilation ($PtcCO_2$ ≥ 50 mmHg during ≥25% of total sleep time). We excluded patients with skin allergy (which is actually not a contraindication of the Sunrise device), patients with a central sleep apnea syndrome and patients at the end of life or with a limitation of active therapies. Children ventilated with a naso-buccal interface and who did not have a smartphone allowing them to install and use the mobile application were excluded.

2.3 | Scoring of PSG

Nocturnal PSGs were performed using the CID102-108D device (Cidelec, Sainte-Gemmes-sur-Loire, France). We recorded respiratory events according to the recommendations of the AASM for children.[20]

PSG recordings were clinically scored by experts from the recruiting centers (Grenoble Alpes University Hospital and Armand Trousseau Hospital, AP-HP, Paris, France). To avoid interscorer variability, PSGs were secondarily anonymized, converted in European data format (EDF), and sent via a secured platform for blinded scoring to the second reference center (Sleep Laboratory, CHU University Catholique of Louvain, Namur, Site Sainte-Elisabeth, Belgium). Scoring was performed according to the recommended

criteria established by the European Respiratory Society (ERS) guidelines.[1,21]

Apneas were defined as an absence or decrease ≥90% of nasal flow for ≥90% of the duration of the event and classified as obstructive, central, or mixed according to the presence or absence of respiratory effort. For obstructive apneas, the duration depends on the child's breathing frequency. For central apneas, they had to be ≥20 s or two respiratory cycles with arousal or at least 3% oxygen desaturation. Hypopneas were scored using the ERS recommended definition of hypopnea, requiring at least a 30% decrease in airflow for more than two or more respiratory cycles and associated with at least a 3% decrease in oxygen saturation measured by pulse oximetry or arousal.[1,21] PSG recordings were analyzed blind to MJM data, and both centers scored PSG recordings independently. OSA was defined according to the ERS guidelines, and OSA severity was classified as mild (AHI < 5/h), moderate (AHI 5–9.9/h), or severe (AHI ≥ 10/h).[1]

2.4 | MJM recordings and description of the Sunrise digital solution

Used to diagnose sleep apnea, the Sunrise digital solution consists of a certified medical device[12,13] that records and analyzes MJM through a wireless single-point of contact sensor placed on the chin (Sunrise, Namur, Belgium). Equipped with an integrated accelerometer and gyroscope, the sensor records the lower jaw's position, rotational movements of the jaw, and its angular velocity within a three-dimensional space at a sampling rate of 25 Hz.

Parents first downloaded the application and then paired the device before attaching the sensor to their child's chin in the mentolabial fold. Recorded data were transferred via Bluetooth Low Energy to a smartphone application during the night. The recorded MJM data were then automatically transferred from the smartphone to a cloud infrastructure at the end of the night (Figure 1). The system then applied a proprietary machine learning algorithm to classify four sleep stages (awake, light/deep, and REM sleep),[22] and respiratory events including obstructive, central apnea/hypopneas, and respiratory effort-related arousal events,[23] based on statistical features extracted from MJM raw signals. The classification outputs are then converted to standard metrics and indices such as the AHI value.

The rhythmic respiratory movements of the mandibular jaw play a crucial role in maintaining pharynx stability during normal sleep. Serving as a dynamic component of craniofacial anatomy, the mandibular jaw actively ensures the patency of the upper airway within the oral cavity. Monitoring these MJM during sleep

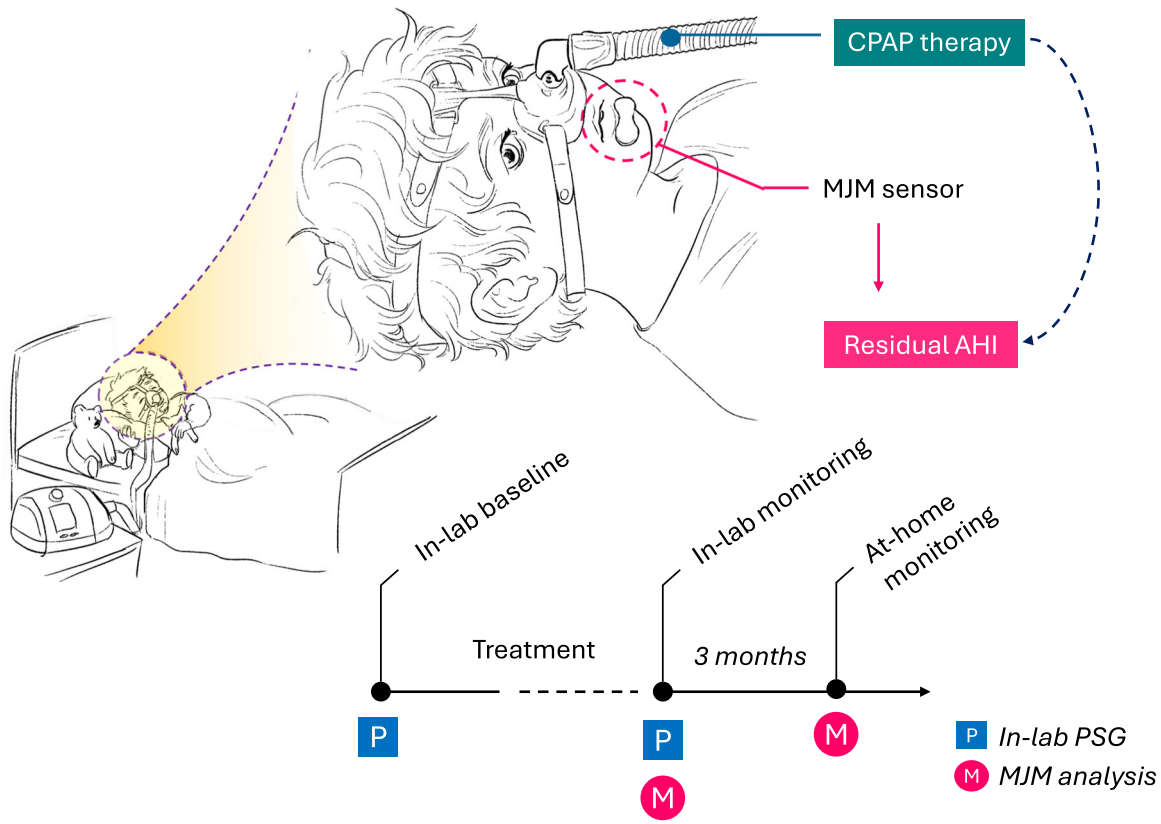

FIGURE 1 Schematic representation of the data recorded by the Sunrise device and the flow chart of the study. CPAP, continuous positive airway pressure; AHI, apnea hypopnea index; MJM, mandibular jaw movements; PSG, polysomnography. [Color figure can be viewed at wileyonlinelibrary.com]

TABLE 1 Characteristics of study population.

Factors	Frequency (*n*) total *N* = 15	Proportion (%)
OSA type		
1	5	33
2	2	13
3	8	53
Ventilatory support		
CPAP	13	87
NIV	2	13
Symptoms at diagnosis		
Loud regular snoring	14	93
Reported breathing effort	14	93
Witnessed apneas	6	40
Nocturnal mouth breathing	5	33
Night sweating	2	13
Nonrestorative sleep	10	67
Daytime sleepiness	10	67
Attentional deficits	2	13
Headaches	2	13

Abbreviations: CPAP, continuous positive airway pressure; NIV, noninvasive ventilation; OSA, obstructive sleep apnea.

enables the precise quantification of minute displacements, typically within the range of a few tenths of a millimeter. These movements are intricately linked with respiratory cycles and are regulated by the respiratory centers.

2.5 | Data analysis

Data analysis and visualization were carried out using R programming language (R Core Team).[24] Data distribution was described as median (5th–95th percentiles range) for continuous variables and frequency (proportion) for categorical variables. The agreement between PSG and MJM monitoring for measuring the residual AHI was evaluated using a Bland–Altman analysis. Median measurement bias and nonparametric percentiles-based limits of agreement (LOA) were reported. A bias-corrected accelerated bootstrap process was used to determine the 95% confidence interval of these results. The average treatment effect of ventilation therapy was determined by evaluating the absolute and relative changes in AHI between baseline (PSG) and posttreatment measurements (in-lab PSG, in-lab MJM recording and one night of MJM monitoring at-home 3 months later). These estimations were based on a generalized linear mixed-model with Gamma distribution. Confidence intervals for the marginal effects were determined by the delta method (marginal effects package). Statistical inferences were based on null hypothesis testing at a significance level of 0.005.[25]

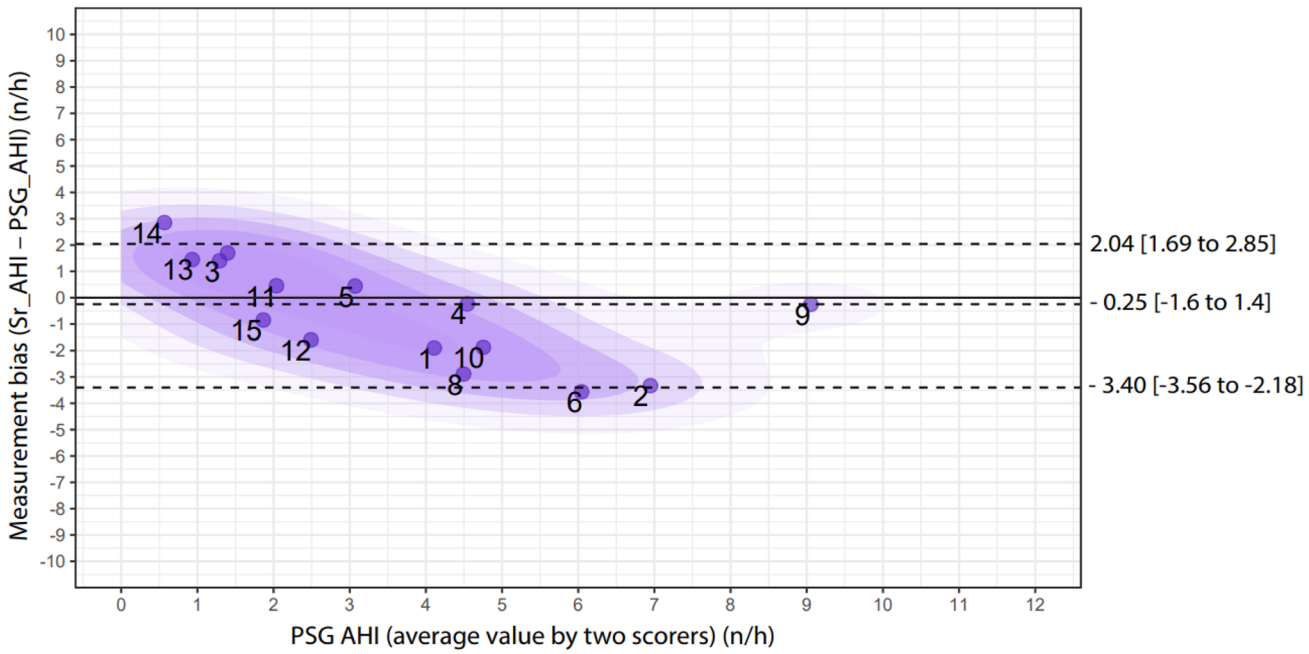

FIGURE 2 Bland–Altman plot evaluating the agreement between in-lab PSG and concomitant MJM analysis for measuring residual AHI in 15 children. The *x*-axis represents the reference scale for AHI estimated by PSG (average value by two scorers), and the *y*-axis represents the scale of measurement bias between MJM analysis and PSG. Each point on the scatter plot represents an individual patient; the three horizontal dotted lines indicate the median value, the upper (95th percentile) and the lower (5th percentile) limits of the measurement bias. AHI, apnea hypopnea index; MJM, mandibular jaw movements; PSG, polysomnography. [Color figure can be viewed at wileyonlinelibrary.com]

3 | RESULTS

3.1 | Characteristics of study population

A total of 15 children (nine boys) were enrolled (eight in center 1 and seven in center 2), with a median age of 12 years, ranging from 5 to 18 years. The weight and body mass index showed a diverse distribution with median values of 49 kg and 23 $kg.m^{-2}$, respectively. The clinical characteristics of the study population are described in Table 1. Before ventilatory support, all participants had OSA as confirmed by PSG, with a median AHI value at baseline of 15.0 events/h (5th–95th percentiles range 8.5–42.6), indicating a high degree of severity. Among the children, eight presented a type 3 OSA (53%). Clinical data revealed a high prevalence of symptoms and signs related to sleep-disordered breathing. Among the children ventilated, 87% were on CPAP (Table 1).

3.2 | Agreement between PSG and MJM analysis in measuring the efficacy of treatment

We first evaluated the validity of residual AHI measured by MJM analysis in comparison with the polysomnographic scoring. The Bland–Altman analysis (Figure 2) indicated a good agreement between the two methods in measuring residual AHI under CPAP/NIV treatment, with a median measurement bias of only −0.25 events/h (95% CI: −1.6 to 1.4) and a LOA of −3.40 to 2.04 events/h which is clinically acceptable (Figure 2).

The distribution of residual AHI under CPAP/NIV treatment evaluated by three different methods (in-lab PSG, in-lab MJM, and at-home MJM analysis) are described in Table 2. Compared with the baseline, there was a consistent and significative improvement in residual AHI under treatment across the three methods, which corresponded to the relative reductions of −83.19% (95% CI: −89.03 to −74.25) for in-lab PSG, −85.18% (−88.74 to −80.5) for in-lab-MJM, and −84.11% (−87.71 to −79.45) for at-home MJM (Table 3). The average and individual trends of AHI change in response to CPAP/NIV treatment per method are visualized in Figure 3.

TABLE 2 Distribution of baseline and residual AHI.

Condition	Method	Median	5th–95th percentiles	*p* Value[a]
Baseline AHI	In-lab PSG	15.00	8.50–42.64	—
Residual AHI	In-lab PSG	3.06	0.83–7.57	<0.001
	In-lab MJM	2.70	0.97–5.65	0.001
	At-home MJM	2.90	0.97–7.50	<0.001

Abbreviations: AHI, apnea hypopnea index; MJM, mandibular jaw movements; PSG, polysomnography.

[a]*p* Value based on Wilcoxon sign-rank test verifying the null hypothesis that there was no difference in AHI score between two conditions.

4 | DISCUSSION

The use of MJM with analysis supported by artificial intelligence showed a good agreement with the PSG-derived AHI in children treated by CPAP or NIV with nasal or nostril masks. Artificial intelligence analysis of MJM therefore holds great promise for at-home monitoring of children under PAP therapies. Our study showed the feasibility and reliability of MJM automated analysis in the laboratory and at home to characterize the OSA burden at diagnosis[13] and under treatment.

Automated MJM-derived AHI measurements have demonstrated comparable performance to in-laboratory PSG in children treated by CPAP or NIV, suggesting that MJM monitoring is an effective and practical means of testing for OSA in the patient's home. There is a growing need for simple, automated tools for the diagnosis and monitoring of OSA that can be used remotely,[26] but it is important that these monitors remain accurate and reliable. Previously, the Sunrise device, using MJM-derived AHI and automated analysis, has been shown to have reliable concordance with in-laboratory-recorded PSG data for the diagnosis of OSA in children with moderate to severe OSA.[13] The benefits of using data recorded at home include a reduction in patient stress associated with travel and overnight stays in hospital, as well as a potential reduction in waiting times and clinical costs. In addition, manual analysis of PSG data is challenging; it is time-consuming, and there is a variability between experts, despite the use of standardized scoring criteria.[27]

This study is the first to compare MJM monitoring with PSG recording in CPAP-NIV treated children. However, several limitations should be highlighted. First, the sample size is limited. Second, the increasing use of technology in healthcare may lead to problems related to lack of access, either due to reduced internet availability in remote areas, lack of familiarity, for

TABLE 3 Change in the average apnea hypopnea index from baseline based on three evaluation methods.

Method of measurement	Absolute change (events/h)		Relative change (%)		
	Estimated	95% CI	Estimated	95% CI	*p* Value
In-lab PSG	−16.67	−23.47 to −9.84	−83.19	−89.03 to −74.25	<0.001
In-lab MJM	−16.53	−22.51 to −10.56	−85.18	−88.74 to −80.50	<0.001
At-home MJM	−16.81	−23.73 to −9.89	−84.11	−87.71 to −79.45	<0.001

Abbreviations: CI, confidence interval; In-lab, in-laboratory; MJM, mandibular jaw movements; PSG, polysomnography.

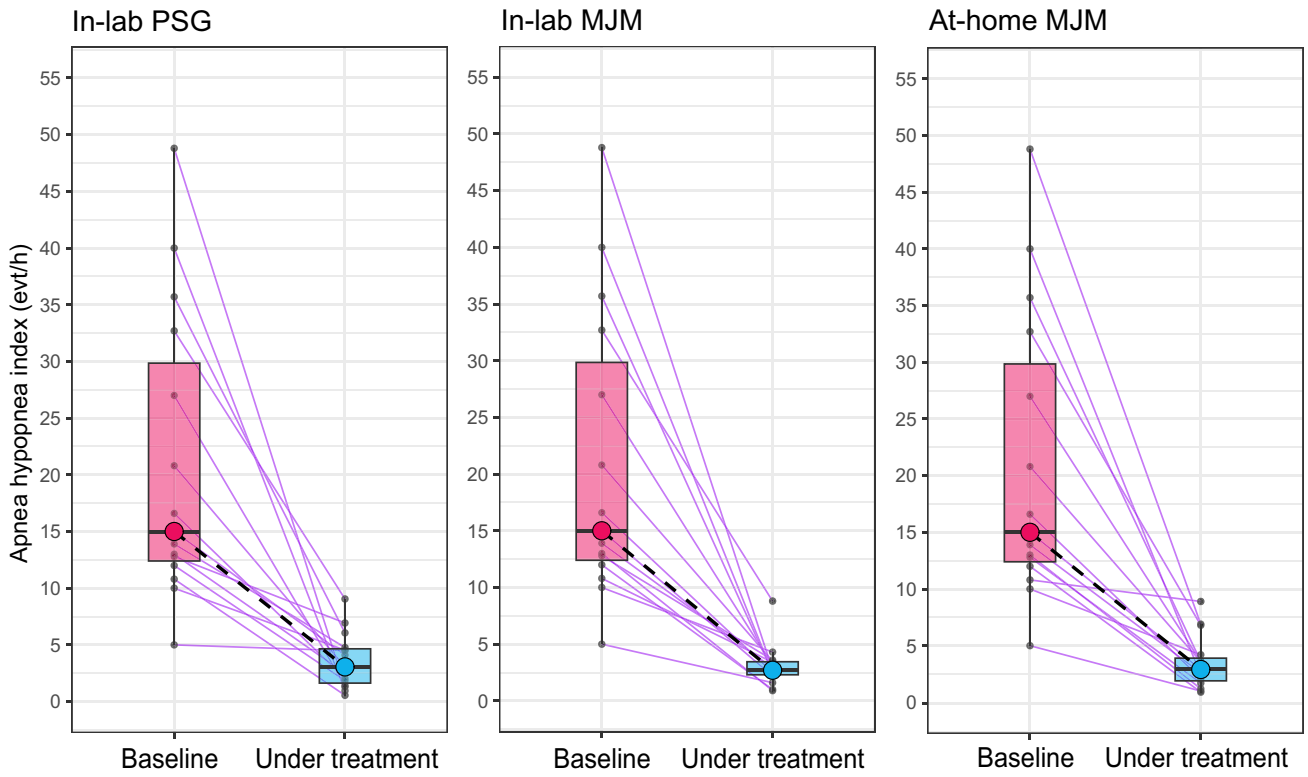

FIGURE 3 Distribution of AHI at baseline and under treatment, based on PSG or MJM analysis. These plots show the tendency of AHI change in response to CPAP/NIV treatment, with the residual AHI measured by three different methods (from left to right): in-lab PSG, in-lab MJM, and at-home MJM. For each plot: the box-plot summarizes the main characteristics of AHI distribution at two time-points, including median, IQR, minimum, and maximum values. The larger dots indicate the median AHI value at each time point, and the dotted line connecting these dots indicates the trend of AHI change at the population level. In the background, a combination of dots and line plots shows individual changes in AHI from baseline to the night of control under treatment. AHI, apnea hypopnea index; CPAP, continuous positive airway pressure; IQR, interquartile range; MJM, mandibular jaw movements; NIV, noninvasive ventilation; PSG, polysomnography. [Color figure can be viewed at wileyonlinelibrary.com]

example, among those who did not use mobile devices when they were younger, or other socioeconomic factors. Improved technology and better access to training could alleviate some of these problems.

5 | CONCLUSION

For future routine clinical practice, MJM with machine learning analysis, which demonstrated a good agreement with manually scored PSG, represents a promising option for monitoring children treated with PAP therapies to optimize their parameters, particularly at home. Given the lack of access to PSG, the time has come to develop these simplified techniques on a large scale for monitoring sleep apnea in children fitted with ventilatory support.[28–30]

AUTHOR CONTRIBUTIONS

Julie Cassibba, Guillaume Aubertin, Jean-Benoit Martinot, Guillaume Mortamet, and Jean-Louis Pépin wrote the manuscript that was reviewed by all the authors. Nam Le Dong, Eglantine Hullo, Nicole Beydon, and Audrey Dupont-Athénor contributed to the data collection and analysis.

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CONFLICT OF INTEREST STATEMENT

The authors declare no conflict of interest.

DATA AVAILABILITY STATEMENT

The data that support the findings of this study are available on request from the corresponding author. The data are not publicly available due to privacy or ethical restrictions.

ETHICS STATEMENT

Written informed consent was obtained from all caregivers.

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