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Clinical validation of a mandibular movement signal based system for the diagnosis of pediatric sleep apnea

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Abstract

Background: Given the high prevalence and risk for outcomes associated with pediatric obstructive sleep apnea (OSA), there is a need for simplified diagnostic approaches. A prospective study in 140 children undergoing in-laboratory polysomnography (PSG) evaluates the accuracy of a recently developed system (Sunrise) to estimate respiratory efforts by monitoring sleep mandibular movements (MM) for the diagnosis of OSA (Sunrise).

Methods: Diagnosis and severity were defined by an obstructive apnea/hypopnea index (OAHI) ≥ 1 (mild), ≥ 5 (moderate), and ≥ 10 events/h (severe). Agreement between PSG and Sunrise was assessed by Bland–Altman method comparing respiratory disturbances hourly index (RDI) (obstructive apneas, hypopneas, and respiratory effort-related arousals) during PSG (PSG_RDI), and Sunrise RDI (Sr_RDI). Performance of Sr_RDI was determined via ROC curves evaluating the device sensitivity and specificity at PSG_OAHI ≥ 1, 5, and 15 events/h.

Results: A median difference of 1.57 events/h, 95% confidence interval: −2.49 to 8.11 was found between Sr_RDI and PSG_RDI. Areas under the ROC curves of Sr_RDI were 0.75 (interquartile range [IQR]: 0.72–0.78), 0.90 (IQR: 0.86–0.92) and 0.95 (IQR: 0.90–0.99) for detecting children with PSG_OAHI ≥ 1, PSG_OAHI ≥ 5, or PSG_ OAHI ≥ 10, respectively.

Conclusion: MM automated analysis shows significant promise to diagnose moderate-to-severe pediatric OSA.

KEYWORDS

machine learning, mandibular movement, pediatric obstructive sleep apnea

1 | INTRODUCTION

Obstructive sleep apnea (OSA) is a highly prevalent condition in children (1%–5%) and is associated with severity-dependent increases in the risk for adverse outcomes, namely neurocognitive and behavioral deficits, and cardiovascular and metabolic morbidities.[1–5] In this context, the American Academy of Pediatrics has recommended in-lab polysomnography (PSG) assessment for children who snore and have at least one other symptom evoking OSA.[6] Unfortunately, PSG is expensive, labor-intensive and cumbersome

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for both parents and children. Owing to a limited number of board-certified pediatricians specialized in sleep medicine such a diagnostic pathway is unrealistic and does not allow for addressing the actual needs thereby leading to significant limitations and delays in access to care.[7] Very few Artificial Intelligence-based approaches have been explored and validated in children at a time that access is also hampered by coronavirus disease 2019 (COVID-19) exposure.

Accordingly, there is a need to adopt alternative methods that will increase accessibility while providing reliable OSA diagnosis in symptomatic children. Prior attempts to achieve such goals have included questionnaires or nocturnal pulse oximetry approaches, which have been fraught with limited success.[8–12] However, American Academy of Sleep Medicine (AASM) does not currently endorse the use of home sleep testing (HST) for diagnosis of OSA in children based on the insufficient scientific evidence regarding HST feasibility and validity. Major concerns about abbreviated diagnostic tests such as HST referred to the inability to detect hypercapnia and of identifying arousals.[13–17] Furthermore, obstructive events, and increases in respiratory efforts with or without arousals may not necessarily be reflected by gas exchange alterations or even by visually recognizable EEG arousals. This is restricting the recognition of respiratory-related sleep fragmentation that has been identified as an important contributor to OSA morbidity.[18–21] Thus, it is highly desirable when adopting innovative techniques for diagnosis of pediatric OSA to include a reliable assessment of respiratory effort (RE). Application of such advanced diagnostic approaches has recently greatly benefited from the advent of machine learning applications in the field of sleep medicine, as those techniques can facilitate complex signal processing and improve reproducibility of sleep staging and respiratory pattern evaluation.[22–25]

It has been previously shown that specific patterns of mandibular movements (MM) can readily identify sleep RE and arousals in an adult population, and that a machine learning framework based on MM automated analysis showed robust diagnostic performance in adults with symptoms suggestive of OSA.[26] In the present study, we aimed to assess the diagnostic capabilities of this novel technology that incorporates MM analysis (Sunrise, Belgium) in a population of consecutive pediatric patients clinically referred for suspicion of OSA and compare it to overnight polysomnographic findings. We therefore hypothesized that the Sunrise-derived "obstructive respiratory disturbance index" (Sr_RDI) would provide satisfactory clinical accuracy to rule-in a diagnosis of pediatric OSA while using AASM severity thresholds.

2 | METHODS

2.1 | Design

This prospective diagnostic study was conducted at CHU UCL – Namur (Belgium). All participants were recruited and scheduled for overnight in-lab PSG according to indications set forth in the pediatric clinical practice guidelines.[6] Written informed consent was obtained from all caregivers. The protocol for this study was approved by the Medical Ethics Committee of the Clinique et Maternite Sainte Elisabeth Namur Belgium (B166201215073). This study followed the Standards for Reporting of Diagnostic Accuracy (STARD) reporting guideline.

2.2 | Participants

Pediatric patients aged 3–17 years referred to the sleep laboratory for clinically suspected OSA (documented history of difficult nocturnal breathing, snoring, witnessed apnea, choking and/or gasping during sleep, night sweat, irritability, restlessness, etc.) were included from September 2017 to November 2019. OSA was suspected on the basis of symptomatology reported by parents. Friedman tongue position classification was reported for all patients as a grading system used to evaluate the relationship of the palate to the tongue.[27] The presence of symptoms led to PSG evaluation after consultation with a sleep specialist. Children with significant, chronic medical conditions, such as genetic syndromes, diabetes mellitus, craniofacial anomalies, or neurologic disease were excluded. Children receiving medications that could affect sleep (sedatives, or systemic corticosteroids) were also excluded. Recordings having incomplete or corrupted data and/or a sleep duration of less than 4 h were excluded.

2.3 | Polysomnography

Routine laboratory based PSGs were recorded with XDream Medatec device (Medatec, Belgium). The parameters monitored included EEG (Fz-A+, Cz-A+, and Pz-A+), right and left electro-oculogram, submental EMG, tibial EMG, chest and abdominal wall motion by respiratory inductance plethysmography (SleepSense S.L.P., USA), nasal and oral flows respectively with a pressure transducer and a thermistor, and O_2 saturation (SpO_2) by digital oximeter displaying pulse waveform (Nonin Medical, USA) enabling the calculation of pulse transit time.

PSG scoring was performed by experienced technician who was blind to the study hypothesis and aims. The PSG data were manually scored in accordance with the recommended criteria in the scoring manual published by the AASM Manual for the Scoring of Sleep and Associated Events and using Domino version 3.0.0.1 software.[28]

The PSG criterion for OSA diagnosis required the presence of one or more obstructive events (obstructive or mixed apnea or obstructive hypopnea) per hour of sleep, coupled with snoring, paradoxical or asynchronous thoracoabdominal movements, or flattening of the inspiratory nasal pressure waveform. End tidal PCO_2 monitoring was not used to support the detection of RE. Increased RE was identified by the presence of a prolonged sequence of breaths characterized by a plateau on the inspiratory portion of the nasal pressure signal and at least one of

the following associated findings: an out of phase in thoracoabdominal excursions, an inspiratory pulse transit time increase up 15 ms between adjacent cardiac beats, or video and microphone confirmed snoring.

2.4 | MM recordings

MM were recorded with the Sunrise system (Namur, Belgium), a coin-sized hardware positioned by the sleep technician on the mentolabial sulcus. The device includes an embedded inertial measurement unit that enables MM sensing and communicates with a smartphone application for external control. The collected MM data were automatically transferred to a cloud-based infrastructure at the end of the night, and data analysis was conducted with a dedicated machine-learning algorithm, trained, and validated in an adult population.[26] Basically, MM result from the muscular activities of the jaw antagonists innervated by the motor branch of the trigeminal nerve.[29] MM are driven by the respiratory centers at the breathing frequency while awake the movement frequency is more variable. In the presence of an upper airway obstruction, MM increase in amplitude in relation with RE mimicking the esophageal pressure swings until arousal closes the mouth and restores the airflow by stiffening and re-opening upper airway (Figure 1). MM analysis allowed for the automatic calculation of an obstructive respiratory disturbance index (Sr_RDI), consisting in the hourly rate of respiratory events marked by RE (obstructive and mixed apneas/hypopneas and respiratory effort-related arousals (RERA). The identification of respiratory disturbances by the Sunrise algorithm relied on the identification of periods of RE characterized by oscillating MM at the breathing frequency and ended by brisk MM of large amplitude indicating the abrupt closure of the mouth, characteristic of arousals or awakenings (Figure 1). Sr_RDI consists of the total number of respiratory disturbances accompanied by RE divided by the total sleep time estimated from the Sunrise analytics.[26]

2.5 | Data analysis

Data analysis was conducted using R statistical programming language.[30] The analysis initially focused on evaluating the agreement between Sr_RDI and PSG_RDI, consisting in the hourly rate of respiratory events marked by RE (obstructive and mixed apneas/hypopneas and RERA). Agreement between Sunrise and PSG was determined using the $ICC_{(3,1)}$ and correlation between the two testing methods was determined using Pearson correlation coefficient (r). Linear regression analysis was performed to characterize the relationship between Sr_RDI and PSG_RDI. Bland and Altman analysis was conducted to estimate the limits of agreement and systematic biases between Sr_RDI and PSG_RDI.[31]

Since clinical diagnosis of pediatric OSA according to the International Classification of Sleep Disorders, Third Edition (ICSD-3) implies the computation of PSG_OAHI (comprising obstructive and mixed apneas/hypopneas and excluding RERA, we optimized the diagnostic performances of Sr_RDI in ruling-in a diagnosis of pediatric OSA at three cut-off thresholds of PSG_OAHI ≥ 1 events/h, PSG_OAHI ≥ 5 events/h, or PSG_ OAHI ≥ 10 events/h. Receiver operating characteristic (ROC) curve analysis was used to evaluate the overall clinical efficacy of the new diagnostic tool via area under the receiver-operating characteristic curve (AUC) and a post hoc analysis to optimize the cutoff points of Sr_RDI for diagnostic decisions compared to the gold-standard cutoff values of obstructive PSG_OAHI recommended in ICSD-3 (1 events/h, 5 events/h, and 10 events/h) was performed. The optimal MM cutoffs were determined at the highest value of the Youden index. The metrics of clinical utility and accuracy were calculated for the defined optimal MM detection cutoff.

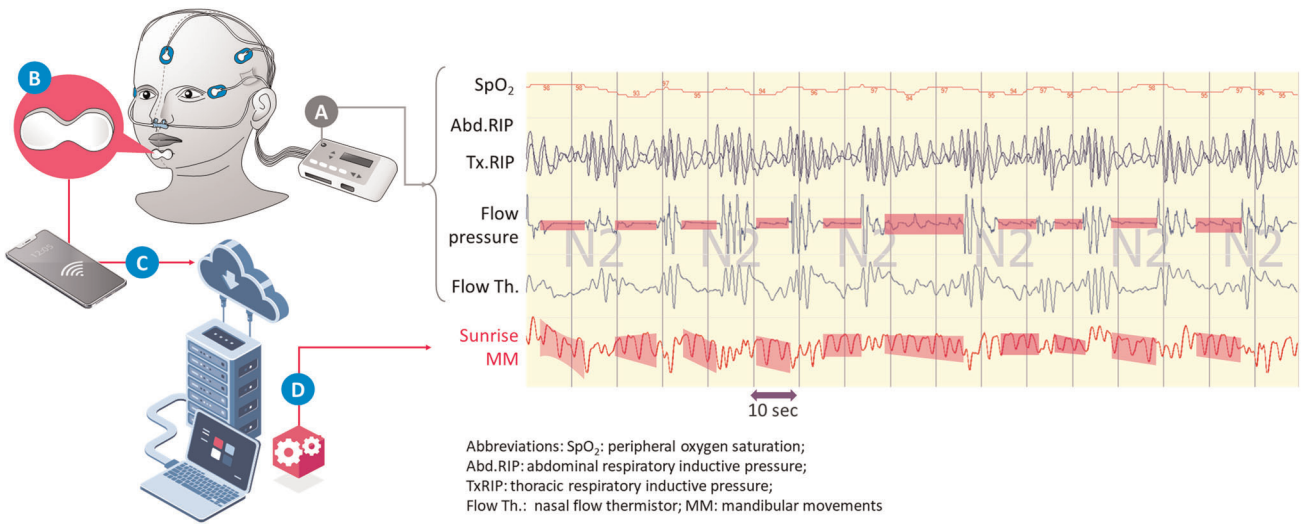

FIGURE 1 Schematic representation of the data concomitantly recorded by in-lab polysomnography (PSG) (A) and Sunrise device (B) but analyzed independently. The data were automatically uploaded into a cloud-based platform (C) and handled by a machine learning algorithm (D). The figure shows the behavior of the Sunrise MM signal after incorporation into PSG fragments marked with episodes of obstructive hypopneas (in red)

3 | RESULTS

3.1 | Characteristics of study population

About 155 patients aged 3–17 years old with clinically suspected OSA completed the sleep study from September 2017 to November 2019. Fifteen patients were excluded for the following reasons: incomplete data (7 patients), total sleep time inferior to 4 h (3 patients), and technical failures (5 patients). The remaining 140 patients were included in the final analysis (mean age: 7.8 ± 4.2 years; mean body mass index: 17.7 ± 5.0 kg/m^2; 55.0% were females). In accordance with the WHO criteria for obesity, 22 children (15.7%) suffered from obesity, and 16 were overweight (11.4%). They were equally distributed across the three PSG identified groups.

The characteristics of the clinical convenience sample are depicted in Table 1.

TABLE 1 Numerical variables are described as median, 5th, 95th centiles and interquartile range

Demographic Characteristics of the Pediatric Cohort n = 140 Sex ratio F/M: 77/63				
	Median	5th centile	95th centile	IQR
Age, years	6.90	3.159	15.841	5.6
Height, cm	121.5	94.9	171.0	36.75
Weight, kg	22.50	13.0	77.4	20
BMI, kg/m^2	16.46	13.89	29.02	3.57
BMI Z_score	0.235	−1.883	2.921	1.84
Neck circumference, cm	17.00	13.29	32.33	5.57
Sleep parameters, PSG				
Total sleep time, h	7.60	4.63	9.00	1.59
OAHI, events/h	0.85	0.01	13.32	2.16
RDI, events/h	5.62	0.94	22.95	5.54
Arousal index, events/h	11.07	5.73	23.65	5.31
Symptoms, complaints and Friedman classification				
Presence of loud snoring	76 (54%)			
Reported breathing effort	56 (40%)			
Witnessed apneas	54 (39%)			
Observed mouth breathing	44 (31%)			
Night sweating	30 (21%)			
Nonrestorative sleep	48 (34%)			
Daytime sleepiness	47 (34%)			
Easily distracted	44 (31%)			
Restlessness	46 (33%)			
Friedman classification	2.28 (0.91)			

Note: Categorical variables were described as frequency (%).
Abbreviations: BMI, body mass index; PSG, polysomnography; OAHI, obstructive apnea/hypopnea index; RDI, obstructive respiratory disturbance index.

3.2 | Association between PSG and MM-derived RDI

Using ICSD-3 criteria, OSA was ruled out in 73 patients (PSG_OAHI < 1 events/h; 52.1%), 47 children (33.6%) were diagnosed with mild OSA, 11 (7.8%) with moderate OSA, and nine (6.4%) with severe OSA.

Figure 2 shows a linear relationship between Sr_RDI and PSG_OAHI in the three clinical groups ($r = .76$, $p < .001$) and the relationship between Sr_RDI and PSG_RDI, including OAHI and RERAs in the three clinical groups. There was a significant correlation between the Sr_RDI and PSG_RDI ($r = .84$; 95% confidence interval [CI]: 0.79–0.89; $p < .001$). PSG_RDI values could be estimated from Sr_RDI by a simple linear regression equation: PSG_RDI = 1.13 * Sr_RDI − 2.67 ($R^2 = .71$; $p < .001$).

3.3 | Intermethods agreement analysis

There was an acceptable agreement between the two methods (PSG vs. Sunrise) in estimating RDI, as suggested by an intraclass correlation coefficient (ICC) of 0.79 (95%CI: 0.72–0.85; $p < .001$).

A Bland–Altman analysis (Figure 3) between Sr_RDI and PSG_RDI showed that the median difference between the two methods was 1.57 events/h with a confidence interval including the zero value and no systematic bias between the two measures. The distribution of the measurement bias within 95% limits of agreements ranged from −2.49 to 8.11. Group-wise analysis highlighted that in the non-OSA group ($n = 73$), the disagreements were normally distributed with a median difference of 2.21 events/h (95%CI: −1.12 to + 8.36). Disagreements were also normally distributed among patients with mild OSA ($n = 47$), with a median difference of 1.18 (95%CI: −1.66 to 7.61). The distribution of disagreement within the groups of patients diagnosed with moderate-to-severe OSA ($n = 20$) was skewed due to outliers at values above 20 events/h on the PSG_RDI scale), with a median difference of −0.06 (95%CI: −16.24 to 5.44).

On the Kernel density plot, the distributions of PSG_RDI and Sr_RDI values represent a high uniformity within each clinical subgroup determined by the conventional cut-off thresholds. The density plots comparison also indicated a large overlap between non-OSA and mild OSA subgroups. These graphs allow expecting the impact of switching technology on the clinical performance of Sr_RDI in OSAS diagnosis, with regard to the reference method (PSG_RDI) in moderate-to-severe OSA children (Figure 4).

3.4 | Diagnostic ability of the MM-derived RDI

ROC curve analysis was performed to evaluate the diagnostic performance of Sr_RDI to identify OSA at the three pre-specified cutoff values

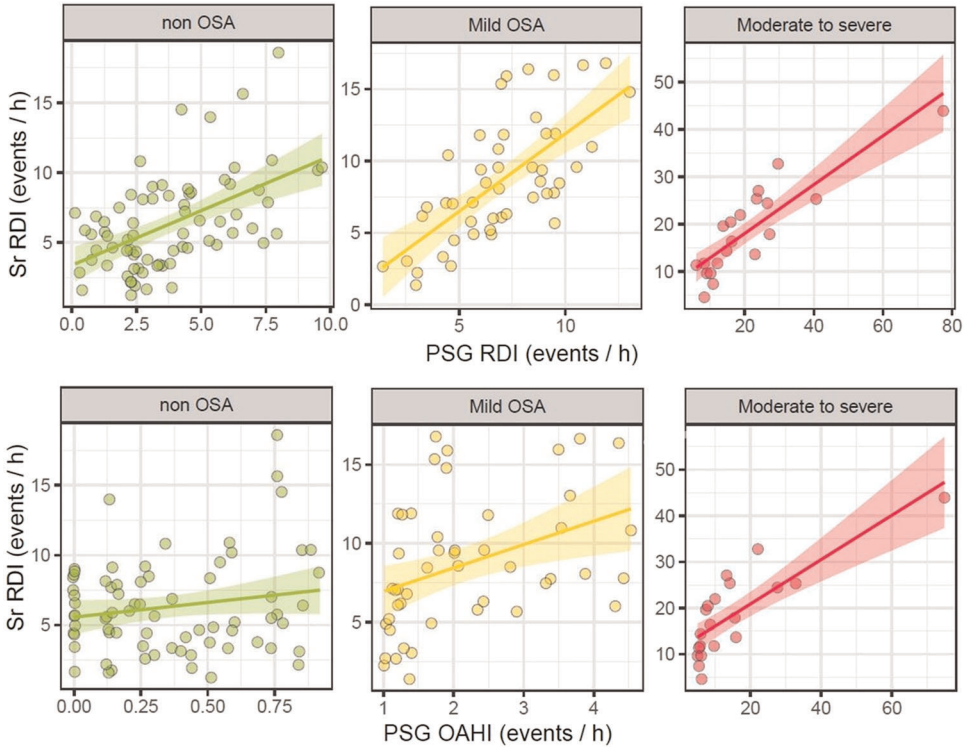

FIGURE 2 Scatter plots describing the relationship between Sr_RDI and PSG_RDI (upper pannel), and Sr_RDI and PSG_OAHI (lower pannel) for non OSA (green), mild OSA (yellow) and moderate-to-severe OSA (red). OSA, obstructive sleep apnea; PSG_OAHI, obstructive apnea/hypopnea index during polysomnography; PSG_RDI, respiratory disturbances hourly index during polysomnography; Sr_RDI, Sunrise-derived obstructive respiratory disturbance index [Color figure can be viewed at wileyonlinelibrary.com]

of PSG_OAHI ≥ 1 events/h, ≥ 5 events/h, and ≥ 10 events/h (Figure 5). The area under the ROC curves (AUC) targeting PSG_OAHI ≥ 1, PSG_OAHI ≥ 5, or PSG_ OAHI ≥ 10 reached 0.75 (95%CI: 0.72–0.78), 0.90 (0.86–0.92), and 0.95 (0.90–0.99), respectively.

After cutoff point optimization, we found that at the best thresholds of 5.75 events/h, 9.60events/h, and 13.07 events/h, Sr_RDI allowed for detection of patients with PSG_OAHI ≥ 1, PSG_ OAHI ≥ 5, or PSG_ OAHI ≥ 10 with accuracy levels of 66%, 85%, and 94%, respectively.

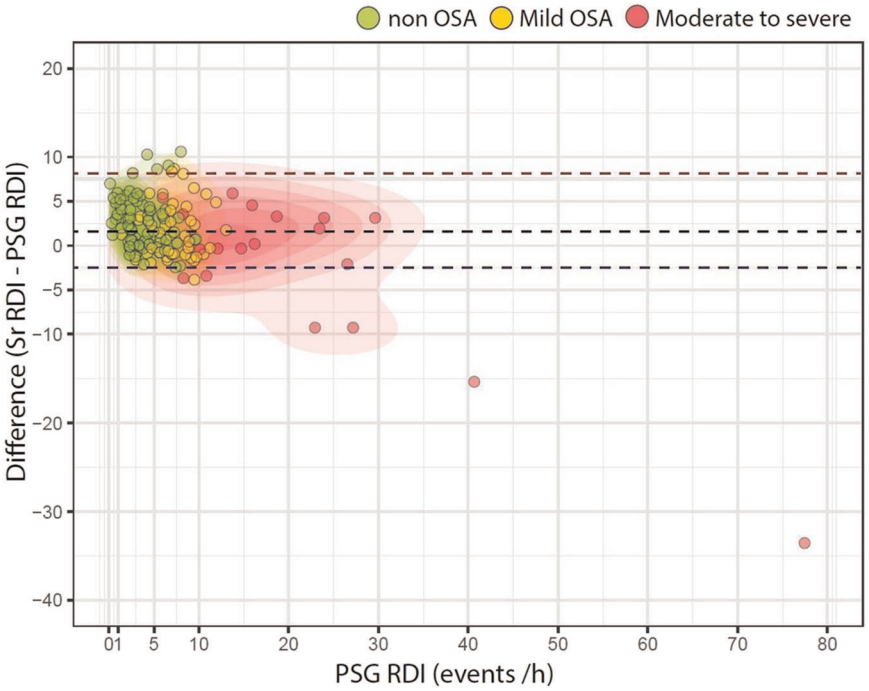

FIGURE 3 Bland–Altman plot between PSG_RDI and Sr_RDI as a function of PSG_RDI, with patients divided into three groups: (1) non-OSA (green), (2) mild OSA (PSG_OAHI ≥ 1; yellow), and (3) moderate-to-severe OSA (PSG_OAHI ≥ 5; red). The horizontal and the dashed lines indicate the median, the 5th and the 95th centiles of the disagreement in the whole sample, respectively. OSA, obstructive sleep apnea; PSG_OAHI, obstructive apnea/hypopnea index during polysomnography; PSG_RDI, respiratory disturbances hourly index during polysomnography; Sr_RDI, Sunrise-derived obstructive respiratory disturbance index [Color figure can be viewed at wileyonlinelibrary.com]

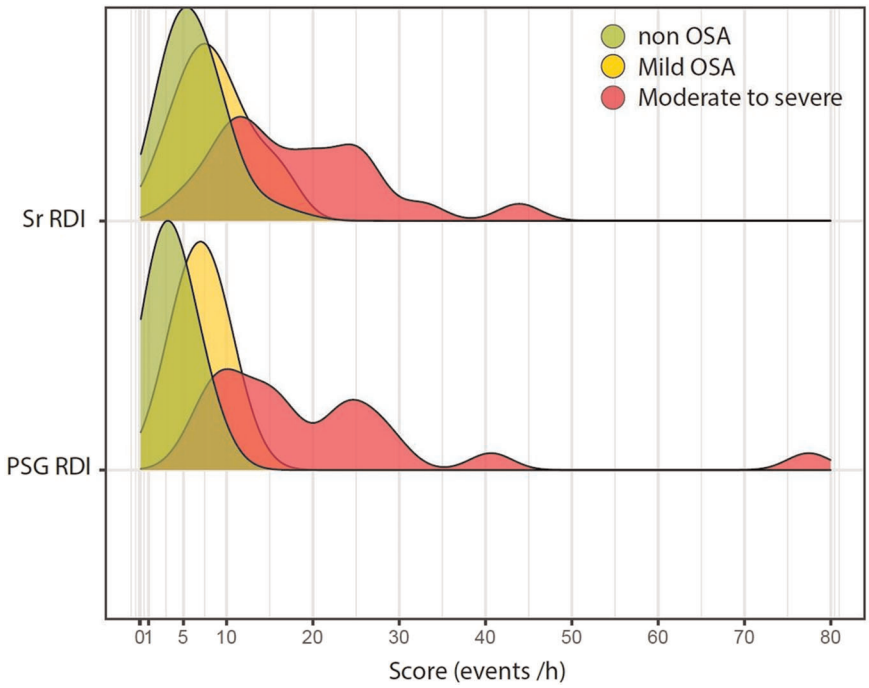

FIGURE 4 Uni-dimensional Kernel density estimations (KDE) plots were to show the true distribution of Sr-RDI and PSG-RDI within the three groups: (1) non-OSA (green), (2) mild OSA (PSG_OAHI ≥ 1; yellow), and (3) moderate-to-severe OSA (PSG_OAHI ≥ 5; red). The KDE plots shared the same scale of RDI. OSA, obstructive sleep apnea; PSG_OAHI, obstructive apnea/hypopnea index during polysomnography; PSG_RDI, respiratory disturbances hourly index during polysomnography; Sr_RDI, Sunrise-derived obstructive respiratory disturbance index [Color figure can be viewed at wileyonlinelibrary.com]

Sr_RDI diagnostic rules allowed us to detect mild OSA with a good sensitivity (83%) but a specificity of 53%, to detect moderate OSA with a balanced sensitivity (90%) and specificity (80%), and to detect severe OSA with an excellent performance (sensitivity of 100% and specificity of 88%; Table 2).

4 | DISCUSSION

In a large prospective pediatric cohort clinically referred for suspected OSA, we evaluated the agreement between MM-derived Sr_RDI and PSG_RDI. The MM simplified diagnosis framework was highly reliable for moderate to severe OSA patients, that is, those clearly requiring treatment interventions. Thus, the current findings reinforce the potential applicability of machine learning derived algorithms coupled with automated analyses for diagnosis of moderate to severe pediatric OSA and should spur renewed efforts for studies in this direction, ultimately leading to wider implementation of such approaches. Our results also demonstrated the capability of this automated analysis to identify RERAs, an element that has clinical significance in children but is too often either ignored or underdiagnosed.[32–34]

PSG_OAHI ≥ 5 in children is a commonly agreed upon criterion for adenotonsillectomy and is associated with an increase in the risk of OSA-associated adverse outcomes.[4,5,17,35] At the best-optimized

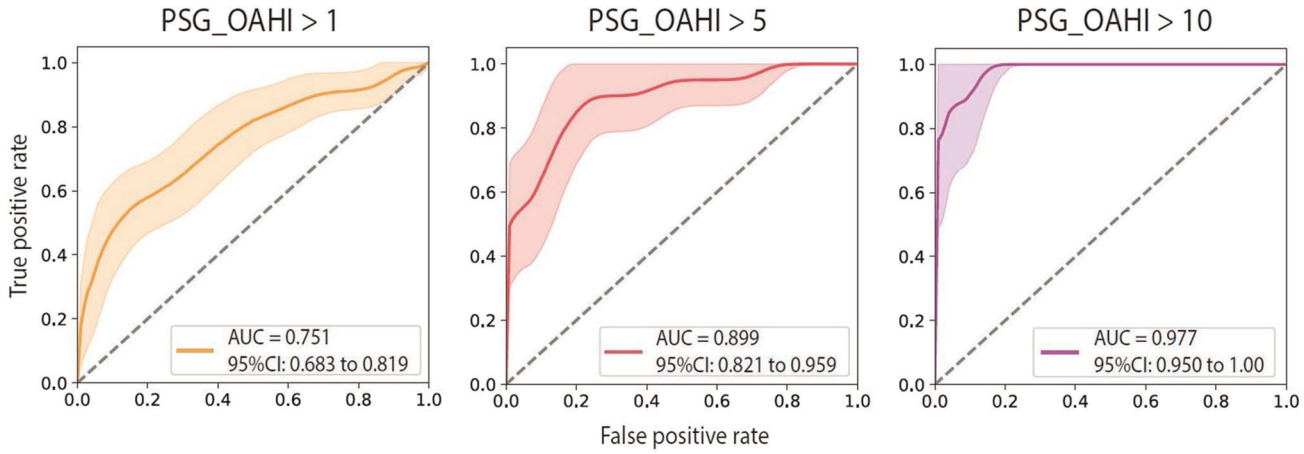

FIGURE 5 ROC curves of the classification rules to detect OSA pediatric patients with PSG_OAHI ≥ 1 (orange curve), PSG_OAHI ≥ 5 (red curve), and PSG_OAHI ≥ 10 (purple curve), from Sr_RDI scores. The smoothing effects on the curves were obtained by bootstrapping. OSA, obstructive sleep apnea; PSG_OAHI, obstructive apnea/hypopnea index during polysomnography; PSG_RDI, respiratory disturbances hourly index during polysomnography; ROC, receiver operating characteristic; Sr_RDI, Sunrise-derived obstructive respiratory disturbance index [Color figure can be viewed at wileyonlinelibrary.com]

TABLE 2 Diagnostic performance of Sr_RDI to detect PSG_OAHI at the diagnostic cutoff values for detecting pediatric OSA

Performance metrics (median and CI)	PSG_AHI≥1		PSG_AHI≥5		PSG_AHI≥10	
Best cut-off	5.75		9.61		13.07	
Sensitivity	0.82	0.78–0.86	0.90	0.87–0.93	1.00	1.00–1.00
Specificity	0.53	0.48–0.59	0.80	0.76–0.84	0.88	0.84–0.91
FPR (false positive rate)	0.18	0.22–0.14	0.10	0.13–0.07	0.00	0.00–0.00
FNR (false negative rate)	0.47	0.52–0.41	0.20	0.24–0.16	0.12	0.09–0.16
PPV (positive predictive value)	0.64	0.59–0.68	0.82	0.78–0.86	0.89	0.86–0.92
NPV (negative predictive value)	0.75	0.70–0.80	0.89	0.85–0.92	1.00	1.00–1.00
F1	0.72	0.68–0.75	0.86	0.83–0.88	0.94	0.93–0.96
BAC	0.68	0.65–0.71	0.85	0.82–0.88	0.94	0.92–0.96
LR + (positive likelihood ratio)	1.76	1.57–2.01	4.52	3.70–5.71	8.28	6.39–11.33
LR-(negative likelihood ratio)	0.33	0.26–0.42	0.12	0.09–0.17	0.00	0.00–0.00
ROC–AUC (area under the ROC curve)	0.751	0.68–0.82	0.90	0.82–0.96	0.98	0.95–1.00

Note: Optimal cutoff points were determined at the highest value of Youden's J index (sensitivity + specificity − 1). The 95% CIs were determined by bootstrapping.

Abbreviations: OSA, obstructive sleep apnea; PSG_AHI, apnea/hypopnea index during polysomnography; PSG_OAHI, obstructive apnea/hypopnea index during polysomnography; PSG_RDI, respiratory disturbances hourly index during polysomnography; ROC, receiver operating characteristic; Sr_RDI, Sunrise-derived obstructive respiratory disturbance index.

cut-off, Sunrise MM driven RDI allowed for the correct identification of patients with PSG_OAHI ≥ 5 events/h with a balanced accuracy of 85% and an area under the receiver operating characteristic curve (ROC AUC) of 0.9. At such level of severity, Sunrise favorably competes with other type four devices incorporating SpO_2 automated analysis for the diagnosis of pediatric OSAS.[11,12,36] Our results show good diagnostic performances to efficiently "rule in" a diagnosis of moderate-to-severe OSAS, the group representing the most critical population to be diagnosed when using simplified approaches. Moreover in the group with PSG_OAHI ≥ 5, the rate of false positive with Sr_RDI was minimal (10%) keeping very low the risk of incorrect diagnosis of OSA potentially treated unnecessarily.

However, the performance of the diagnostic algorithm for detecting OSA from non OSA patients using the clinical threshold of PSG_OAHI ≥ 1 events/h was less optimal. About 73 habitually snoring symptomatic children referred by their primary care physicians for clinical suspicion of OSAS showed OAHI < 1 event/h in the PSG.

The Sr_RDI cut-off of 5.75 as selected would misclassify 18% of patients as false positives and 47% as false negatives. We should emphasize that this cutoff of 1 event/h will pose difficulties irrespective of the diagnostic approach due to the very low signal-to-noise ratio imposed by this cutoff. One could therefore argue that the conventional rules to reach a diagnosis of pediatric OSA should be modified to incorporate RERAs such as to improve the concordance between PSG-derived indices, complaints/symptoms, and outcomes.[31,37] It has been suggested that RERAs and snoring may better predict both cognitive and behavioral problems in young children than the commonly used PSG_OAHI.[37–39] The high incidence of RERAs in children is further highlighted by the PSG_RDI, which added an average of 4.25 events/h (*SD*: 2.61 events/h) to the PSG-OAHI. MM-based technology is designed to identify the presence of these additional RERAs. The current recommended cut-off is probably not the best predictor of deleterious outcomes, since it was chosen on the basis of the statistical distribution of normative PSG data rather than drawn from considerations of pathophysiological inputs.[40,41] In addition, the "gold standard" PSG is known to vary from one night to the other depending on sleep architecture and body position, especially at the lower end of the severity spectrum. This could also lead to imprecisions in the diagnosis after a single night in the laboratory, particularly when applying the 1 event/h cutoff. In light of the readily available accessibility and scalability, along with the associated low cost and automatic scoring supported by machine learning, MM analysis could offer a valid option for implementation of OSA diagnosis at home over several nights, thereby improving the reliability of the test and better reflecting the actual disease burden in any given symptomatic child.

As mentioned, there have been multiple alternatives to PSG that have been examined over the last three decades in an effort to improve the accessibility of symptomatic children to a timely diagnosis and treatment. These options have ranged from questionnaires with limited diagnostic accuracy precluding their use as a routine diagnostic tool for OSA to more promising approaches relying on automated machine learning-based analysis of single-channel SpO_2.[8,9,11,12] The technique described here positively contributes to this effort by presenting for the first time the diagnostic accuracy of a mandibular movements-based system in confirming or discarding the presence of OSA in symptomatic

children. The combination of MM monitoring with machine learning analysis has been recently proven as efficient for OSA diagnosis in adults.[26] Similarly, investigation of MM patterns after adenotonsillectomy in children provided compelling evidence as to the value of monitoring RE in a pediatric population, since MM patterns during sleep inform about the changes in respiratory drive in the presence of an increase in upper airway resistance.[42,43] Thus, the proposed novel technique is of considerable interest, as it has the potential to simplify the process of pediatric OSAS diagnosis, to unclog sleep centers and orient pediatric OSA diagnosis to home-based settings, and to prevent from COVID-19 exposure risk. This brings obvious advantages for both children and their caregivers.[44] Other technologies that use multiple channels in the home remain very labor intensive and require expert scoring and are fraught with high rates of technical failures. The mandibular jaw movement-sensing hardware could be in the future complemented by a pulsed oximetry system informing about the risk of respiratory effort related oxygen desaturation. Indeed, synchronization with oximetry would potentially increase the specificity of the technique. The Sunrise solution is currently available in the market, and is in the process of receiving approval by several national health agencies.

4.1 | Study limitations

The implementation of such a solution in ambulatory settings will need additional validation to thoroughly evaluate its diagnostic accuracy in unattended home settings. Ideally, future studies will be multicentric, and therefore allow for generalization of the present results. A larger sample size is also required in further studies for more robust representation of the OSAS spectrum of disease. Another limitation was that 10% of patients were excluded due to problems related to missing data in the questionnaires or in the polysomnography (signal loss). Technical failures related to the connected Sunrise system were due to the loss of the wireless connection, and were observed in three children from the group discarded from the final analysis. Finally, further studies might examine the potential improvement of the Sunrise system performance by incorporating pulse oximetry in the diagnosis of OSAS.

5 | CONCLUSIONS

A machine-learning approach based on mandibular movement analysis displays acceptable accuracy as a tool for the diagnosis of moderate-to-severe OSA in symptomatic children. This approach may provide a suitable and convenient home-based alternative for the assessment of pediatric OSA in the near future.

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CONFLICT OF INTERESTS

The study was performed at CHU UCL Namur Site Sainte-Elisabeth (Belgium). The devices used in the study were provided by Sunrise (Namur, Belgium). Dr. Pépin is a scientific advisor of Sunrise; he also reports grants and personal fees from ResMed, grants and personal fees from Philips, grants from Fisher and Paykel, grants and personal fees from Sefam, grants from Astra-Zeneca, grants and personal fees from Agiradom, personal fees from Elevie, grants and personal fees from Vitalaire, personal fees from Boehringer, outside the submitted work. Dr. Martinot is a scientific advisor of Sunrise; he is a remunerated investigator in Pharma trials for Jazz Pharmaceuticals and Theranexus, outside the submitted work. The other authors have no conflicts of interest to disclose linked to this study or direct involvement in the development of the device used here to measure mandibular movements. The other authors received no external funding.

CONSENT STATEMENT

Written informed consent was obtained from all caregivers.

AUTHOR CONTRIBUTIONS

Jean-Benoit Martinot: conceptualization (lead); data curation (lead); formal analysis (lead); investigation (lead); methodology (lead); supervision (lead); validation (lead); visualization (lead); writing original draft (lead); writing review & editing (lead). **Valérie Cuthbert**: data curation (lead); formal analysis (lead); visualization (lead). **Nathalie Coumans**: data curation (lead); formal analysis (lead); visualization (lead). **Deborah De Marneffe**: data curation (equal); supervision (lead); visualization (lead). **Clément Letesson**: conceptualization (supporting); data curation (supporting); formal analysis (lead). **Jean Louis Pepin**: conceptualization (lead); funding acquisition (equal); supervision (supporting); visualization (supporting); writing original draft (lead); writing review & editing (lead). **David Gozal**: conceptualization (lead); funding acquisition (lead); methodology (lead); supervision (supporting); writing original draft (lead); writing review & editing (lead).

DATA AVAILABILITY STATEMENT

The data sets generated during and/or analyzed during the current study are available from the corresponding author on reasonable request. However, source data for figures [numbers] are provided within the paper.

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