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Original article

Respiratory polygraphy in children: Feasibility in everyday practice in an ENT department and value of automatic detection of respiratory events



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ABSTRACT

Objectives: Using respiratory polygraphy (RP) in children for diagnosis of obstructive sleep apnea/hypopnea syndrome (OSAHS) can be challenging in terms of device acceptance and sensor displacement. Automatic analysis of respiratory events has never been evaluated in a pediatric population. The primary objective of this study was to determine the feasibility of pediatric RP in routine ENT department practice. The secondary objective was to evaluate the reliability of the automatic detection of obstructive and central respiratory events in children.

Methods: A single-center retrospective study included 50 children (32 boys, 18 girls; mean age 5.5 ± 2.3 years) undergoing overnight RP in an ENT department between January and August 2016 for suspected OSAHS. Manual detection of respiratory events was performed by one ENT specialist experienced in RP interpretation, and compared to automatic analysis.

Results: The device was well accepted in 98% of cases. Overall signal quality was $>50\%$ in 76% of cases, with average signal quality of 70.8% (86% in patients >3 yrs, 25% in patients <3 yrs, $P=0.0013$). There was no significant correlation between manual and automatic analyses, except for central apnea (Spearman coefficient 0.43; $P=0.0015$). One hundred percent of patients presented OSAHS according to automatic detection, compared to 32% according to manual detection ($P<0.005$).

Conclusions: Pediatric RP is feasible in routine practice in an ENT department, with good acceptance and satisfactory signal quality in children older than 3 years. Automatic analysis of respiratory events in children is unreliable, except for central apnea.

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1. Introduction

Respiratory polygraphy (RP) consists in sleep recording with simultaneous examination of blood oxygen saturation on oximetry, oronasal airflow, and/or snoring quantification by recording of tracheal noise, and/or respiratory effort detection, and/or body position analysis, for at least 6 hours overnight (according to the French Common Classification of Medical Acts (CCAM) version 55.50, of January 01, 2019). In France, it is currently considered as a good alternative to nocturnal polysomnography (PSG) in a sleep laboratory for diagnosis of childhood obstructive sleep apnea/hypopnea syndrome (OSAHS) [1]. The main advantages are

easier access, simpler sensor setup, shorter interpretation time, and lower cost [2]. Several studies have assessed the correlation between apnea/hypopnea index (AHI) on PSG and RP [3–9], with varying results; most, however, concluded that RP was reliable in moderate to severe OSAHS (AHI >5) [5,7].

Despite being easier to setup than PSG in children, implementation can still be technically challenging in terms of device acceptance, recording quality impairment by sensor displacement, and surveillance modalities [2,10]. In the various studies of the subject, RP was performed either in a sleep laboratory [3–7,9] or at home [3,4,8–13]. The interest of RP in a hospital context but outside of a sleep laboratory is to improve access and implementation time while still having nightlong nursing surveillance. There has been just 1 study including this RP context, with 26 out of 101 patients, the others undergoing RP at home; 88.4% of recordings were interpretable [14]. In adults, recordings are usually read by a relatively reliable automatic signal analysis with systematic

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manual correction. In children, dedicated pediatric automatic analysis is available in most software packages, but reliability has never been specifically assessed.

The main objective of the present study was to assess the feasibility of pediatric RP in the routine practice of a hospital ENT department not specializing in sleep disorder. The secondary objective was to assess the reliability of the manufacturers' software for automatic detection of respiratory events: obstructive apnea/hypopnea and central apnea.

2. Material and method

A single-center retrospective study included 54 consecutive children undergoing RP in the ENT Department of the Montpellier University Hospital Center (France) between January 7 and August 26, 2016. Inclusion criteria comprised: RP for suspected OSAHS, and age 2–12 years at examination. Four patients were excluded for missing clinical data (followed up elsewhere). Analysis concerned 50 files.

Mean age was 5.5 ± 2.3 years (range: 2–11 years; 32 boys, 18 girls). Thirty-two percent ($n = 16$) presented particular diatheses: polymalformative syndrome ($n = 6$), Downs syndrome ($n = 3$), overweight or obesity ($n = 3$), neurologic pathology ($n = 4$).

Indications for RP, following the 2012 French health authority guidelines [9], comprised:

- non-contributive interview (28%, $n = 14$);
- discordance between interview and physical examination (22%, $n = 11$);
- confirmation of diagnosis of suspected type II (overweight) or type III (neurologic, malformative or genetic pathology with craniofacial deformity and neuromuscular or skeletal involvement) OSAHS on the Capdevila-Gozal classification [15] (32%, $n = 16$);
- confirmation of diagnosis in a context of high surgical risk (hemostasis disorder or risk of velar decompensation with velar anatomic abnormality) (4%, $n = 2$);
- postoperative control for suspected residual OSAHS (14%, $n = 7$).

On initial clinical assessment, 74% of parents ($n = 37$) reported nighttime snoring, 16% ($n = 8$) reported definite nocturnal apnea, and 38% ($n = 19$) reported uncertain nocturnal apnea. Fifty-eight percent of children ($n = 29$) had grade 3 or 4, 10% ($n = 5$) grade 2, and 16% ($n = 8$) de grade 1 tonsillar hypertrophy on the Brodsky classification. Flexible nasal endoscopy, performed in 22 children, found adenoid hypertrophy in half of the cases. Sixteen diagnoses of OSAHS were made on RP (32%): 10 type I, 1 type II and 5 type III. Table 1 shows the main data from the 50 polygraphies.

The equipment was a Nox T3 respiratory polygraph (Nox Medical Inc. Reykjavik, Iceland, distributed in France by Resmed, San Diego, CA, USA) [16]. Sensors comprised: a finger sensor to record heart rate, oxygen saturation and pulse photoplethysmogram,

Table 1
Results of respiratory polygraphies.

AHI	0.9 (0.5–2.9)	
Median (1st–3rd quartile)		
Overall signal quality	70.8% (50.8–99.2)	
Median (1st–3rd quartile)		
Signal quality > 50%	76%	$n = 38$
Signal quality > 75%	46%	$n = 27$
Diagnosis after RP		
OSAHS	32%	$n = 16$
Central SAS	8%	$n = 4$
No SAS	58%	$n = 29$
Non-interpretable	2%	$n = 1$

AHI: apnea/hypopnea index; RP: respiratory polygraphy; OSAHS: obstructive sleep apnea/hypopnea syndrome; SAS: sleep apnea syndrome.

thoracic and abdominal inductance belts to measure respiratory effort, and a nasal cannula to assess respiratory flow. Actimetry, body position monitoring and snoring sound recording used sensors included in the central monitor on the thorax.

Sensors were positioned by an ENT specialist with training in childhood sleep disorder. Device acceptance was defined as patient compliance with application and maintenance of the sensors throughout the night, regardless of any involuntary displacements, these being analyzed separately. Such involuntary displacement was checked on the department's usual monitoring protocol: a nurse, not specializing in sleep study but who had been instructed on how to check and if necessary reposition the sensors, visited the child every 3 hours. Programmed recording time was in all cases 8 hours.

RPs were interpreted by a single senior ENT specialist with training in childhood sleep disorder, aware of the clinical file. This manual analysis followed the guidelines of the American Academy of Sleep Medicine (AASM) 2012 [17]. OSAHS was defined by obstructive apnea index (OAI) > 1/hour or obstructive apnea/hypopnea index (OAHl) > 1.5/hour; central apnea syndrome was defined by central apnea index (CAI) > 1/hour [1,18]. Mixed apnea was categorized with obstructive apnea. The automatic analyses by the dedicated Noxturnal software (Nox Medical Inc., Reykjavik, Iceland) were also included in the medical file. Subsequent medical management was founded on the manual analysis only, which counted as gold standard.

Overall recording quality was determined automatically by the software according to presence of artifacts on: nasal airflow, thoracic movements, abdominal movements, and finger sensor signals (oximetry and heart rate). Nasal airflow and thoracic and abdominal movements were considered to be artifacted in case of > 120 s signal absence. Oximetry and heart rate were considered to be artifacted in case of $SpO_2 < 50\%$, pulse < 25 bpm or if the sensor was not in position on the patient's finger. Artifacted signal duration as a function of total analysis time gave signal quality expressed as a percentage for each of the 4 sensors; overall recording quality corresponded to the poorest of these 4 percentage values. Recordings were considered technically acceptable for overall quality > 50%. In case of overall quality < 75%, RP traces were checked to identify the implicated sensor(s). Correlations were assessed between overall quality and RP indications and patient age.

Manual and automatic analyses were compared for respiratory events (total, obstructive apnea, obstructive hypopnea, obstructive apnea + hypopnea, central apnea) as numbers and indices.

The Fisher exact test was used to correlate overall recording quality with RP indications and with age > 3 years. The Student *t*-test was used to correlate recording quality and patient age, and the Pearson χ^2 test to correlate recording quality with age > 5 years. The Wilcoxon signed ranks test was used to compare automatic versus manual analysis, and the Spearman correlation coefficient was used to analyze central apnea.

The study had local review board approval from the Montpellier University Hospital research ethics committee.

3. Results

3.1. Examination feasibility and predictive factors for quality

RP was feasible in 98% of cases ($n = 49$) overall: 100% of under-3 year-olds ($n = 8$), and 97.8% of over-3 year-olds ($n = 42$). The single failure was due to poor acceptance in a child aged 4 years 3 months, with no particular comorbidity. All other children were hooked up easily, with interpretable RP.

Mean overall recording quality was 70.8%, with 76% ($n = 38$) acceptable RPs (overall quality > 50%). RPs with quality < 75%

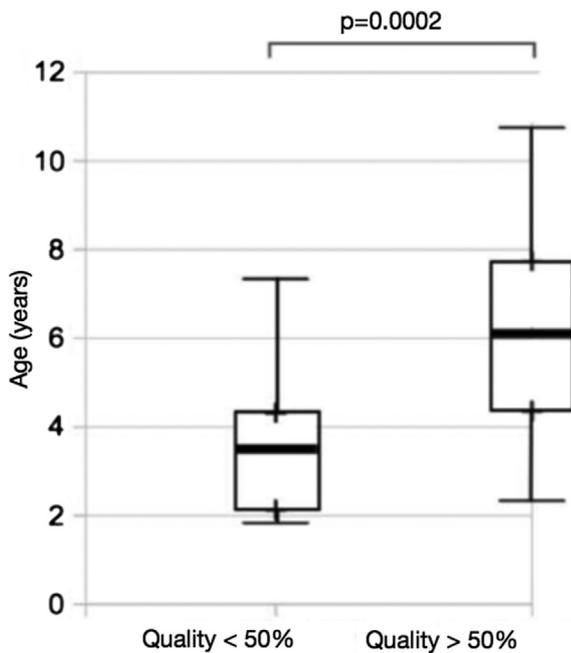


Fig. 1. Correlation between age $>/<$ 3 years and overall examination quality (median, 1st and 3rd quartile, range).

($n = 23$) implicated isolated nasal sensor displacement in 52.2% of cases ($n = 12$), isolated finger sensor displacement in 21.7% ($n = 5$), and displacement of both in 26.1% ($n = 6$).

Indications for RP did not correlate with overall examination quality: $P > 0.05$ for all 5 types of indication. Quality did, however, correlate with age: under-5 year-olds had mean quality $> 50\%$ in 58% of cases ($n = 14/24$), versus 92% ($n = 24/26$) in over-5 year-olds ($P = 0.0046$); a similar difference was found for $<$ vs. $>$ 3 years of age: 25% ($n = 2/8$) versus 86% ($n = 36/42$), respectively ($P = 0.0013$) (Fig. 1).

3.2. Automatic analysis

Total number of respiratory events, obstructive respiratory events (apnea and hypopnea), isolated obstructive apnea, isolated obstructive hypopnea and respective time indices differed significantly between automatic and gold standard manual analyses (Fig. 2). Table 2 shows results on the 2 types of analysis. All patients showed OSAHS on automatic analysis, versus 32% on manual analysis ($P < 0.005$). For central apnea, automatic and manual results were comparable (Fig. 3): Spearman correlation coefficient, 0.43 ($P = 0.0015$) for CAI. For a pathologic CAI threshold of 1, the specificity of automatic analysis was 100% ($n = 45/45$), and sensitivity 50% ($n = 2/4$). In the first of these 2 cases, pathologic CAI at 1.6 went undetected, in a child aged 33 months at examination; overall signal quality here was poor (4.6%), with displacement of both finger and nasal sensors. In the second, a child aged 4 years with no particular history showed CAI = 2.6; device acceptance here was good, and overall quality was 99.7%.

4. Discussion

The gold standard exploration of sleep-disordered breathing (SDB) in children is type-1 nocturnal PSG in a sleep laboratory. RP is classified as type 3 according to the American Sleep Disorders Association (ASDA) (later, American Association of Sleep Medicine: AASM), with the following criteria: ≥ 4 signals, including airflow and thoracoabdominal respiratory effort and electrocardiogram (which may be replaced by heart rate), plus, optionally,

body position, but not electroencephalogram, electro-oculogram or electromyogram, and without mandatory technical in-hospital surveillance [19]. It is accepted by the French Health authority that PSG can be replaced by RP for exploration of obstructive sleep respiratory disorder in children when performed and interpreted by a specialized team [20]. Indications are thus comparable to those for PSG, except for initiation of non-invasive ventilation by continuous positive airway pressure (CPAP) [1,20]. RP has several advantages over PSG. There are fewer sensors, which saves setup time and improves device acceptance. Interpretation is faster, at 20–30 min per examination compared to at least 1 hour for PSG. It is also more accessible than type-1 PSG, not requiring overnight laboratory stay, although this point does not apply to type-2 PSG, implemented at home. And finally, costs are lower, at €145.92 euros (French code GLQP007) versus €214.27 for PSG (code AMQP013) on the French Common Medical Acts Classification (CCAM version 55.50, of January 01, 2019). It has, however, important limitations: non-assessment of total sleep time, and neglect of events causing arousal without desaturation, with risk of false negatives by underestimating the AHI [5]. For a pathologic AHI of 1/hour, Scalzitti et al. reported 81% sensitivity and 60% specificity for RP conducted in a sleep laboratory [4]. Other studies comparing AHI on PSG versus RP, in sleep lab or at home, found greater reliability with an AHI threshold > 3 /hour for OSAHS: sensitivity 90.9% and specificity 94.1% for Alonso-Alvarez et al. [3]; for a threshold of > 5 /hour, sensitivity and specificity were respectively 88% and 98% for Rosen et al. [8], and both 100% for Jacob et al. [9]. It would thus seem that RP loses reliability in mild OSAHS (AHI < 5).

In assessing the feasibility of RP in children, device acceptance needs to be distinguished from recording reliability (technical acceptability) in terms of artifact-free signal duration. The high 98% device acceptance rate matched the literature: 31 out of 33 patients (93.9%) for Scalzitti et al. [4] and 50 out of 50 (100%) for Alonso-Alvarez et al. [3]. For recording reliability, there is no consensus as to what is technically acceptable [2]: 4 hours' artifact-free recording for Brockmann [14], Moss [11] and Rosen [8], $< 40\%$ artifacted airflow over total recording time for Alonso-Alvarez [3], or interpretable data from 3 channels for ≥ 6 hours for Scalzitti [4] and Poels [12]. In view of the above, we set a reliability threshold of 50% overall signal quality for 8 hours of recording; for the 11 RPs with overall quality $< 50\%$, interpretation was still possible, but conclusions were drawn only from interpretable signals and the patient's clinician was systematically advised to repeat recording. The present 76% reliability was comparable to the 75% rate (25 patients out of 33) reported by Scalzitti et al. [4]. Brockmann et al. [14] had a higher rate, at 88.4% (23 out of 26). Although age did not seem to affect device acceptance, recording quality was significantly lower in younger children, due to more frequent sensor displacement during the night, with a particular threshold at 3 years of age. A recent literature review showed that the sensor most liable to get displaced was the nasal cannula, as in the present series [2]. Taking only over-5 year-olds, mean recording quality was excellent, at 92%, confirming the feasibility of RP in older children. This was in agreement with Scalzitti et al., who reported better correlation of RP with PSG results in over-6 year-olds, and suggested restricting RP to this age-group [4]; however, given the OSAHS incidence peak between 2 and 5 years, this would seem unsuited to the needs of paraclinical exploration of OSAHS in children [21]. It is also noteworthy that indications for RP, including suspected type-3 OSAHS ($n = 13$ in the present series) showed no association with overall recording quality; nevertheless, the French Society of ENT recommends PSG in this category of patients [1].

Like in type-2 PSG, the main advantages of RP conducted at home compared to in-hospital recording lie in cost-saving, ease of access, and the more familiar environment, improving sleep quality [9]. There is, however, the problem of sensor displacement, to be

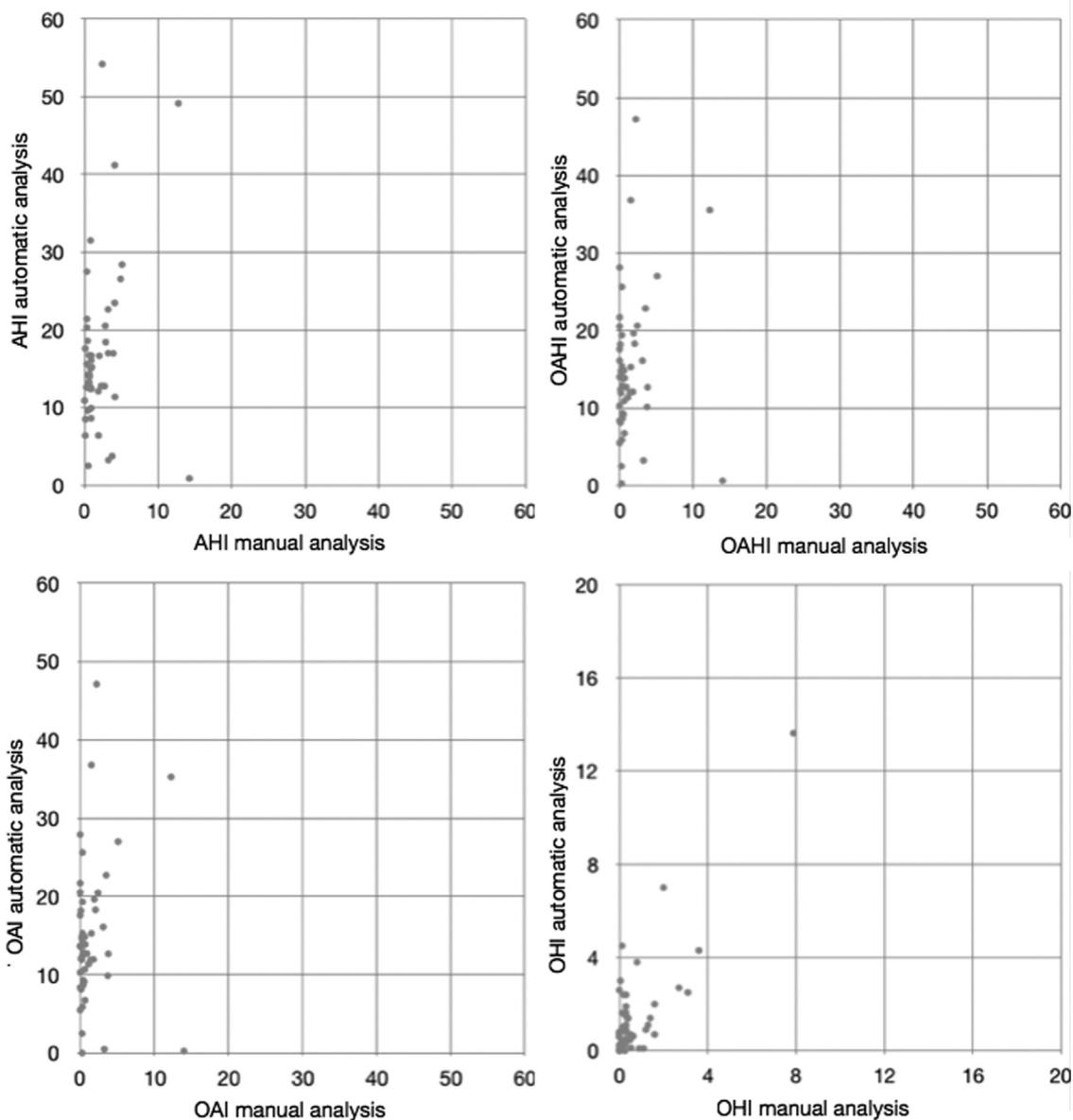


Fig. 2. Apnea/hypopnea index (AHI), obstructive apnea/hypopnea index (OAH), obstructive apnea index (OAI) and obstructive hypopnea index (OHI) on automatic detection according to manual values.

Table 2
Results of manual and automatic detection of respiratory events.

	Manual analysisMedian (1st–3rd quartile)	Automatic analysisMedian (1st–3rd quartile)	P
Number of respiratory events	7.0 (4–20.8)	116 (91.3–143.5)	<0.0005
AHI	0.9 (0.5–2.9)	14.6 (11.6–18.5)	<0.0005
Number of obstructive apneas and hypopneas	3.0 (1.3–11.8)	102.5 (75.3–142.0)	<0.0005
OAH	0.4 (0.2–1.7)	13.3 (9.5–18.3)	<0.0005
Number of obstructive apneas	0.5 (0–2.8)	102.5 (75.3–142.0)	<0.0005
OAI	0.1 (0–0.4)	13.2 (9.4–18.3)	<0.0005
Number of obstructive hypopneas	2.0 (1.0–5.0)	6.5 (3.0–14.5)	<0.0005
OHI	0.3 (0.1–0.8)	0.9 (0.4–1.8)	<0.0005
Number of central apneas	2.0 (1.0–5.0)	1.0 (0–2.0)	<0.0005
CAI	0.3 (0.1–0.6)	0.1 (0–0.3)	<0.0005

AHI: apnea/hypopnea index; OAH: obstructive apnea/hypopnea index; OAI: obstructive apnea index; OHI: obstructive hypopnea index; CAI: central apnea index.

managed by parents who are not trained in the use of the equipment and which may be a cause of anxiety for them. To get around this without loss of accessibility, we opted for RP conducted in hospital but not in a dedicated sleep laboratory. However, comparing the present reliability with reports of RP conducted at home shows

no obvious superiority for in-hospital examination [3,4,8–11,13]. One study, by Poels et al., in 24 children, reported a particularly low rate of 29% [12]; this may have been due to the equipment being fitted by a nurse without training in sleep disorder, which was also the case for Scalzitti et al. [4], although their reliability rate was

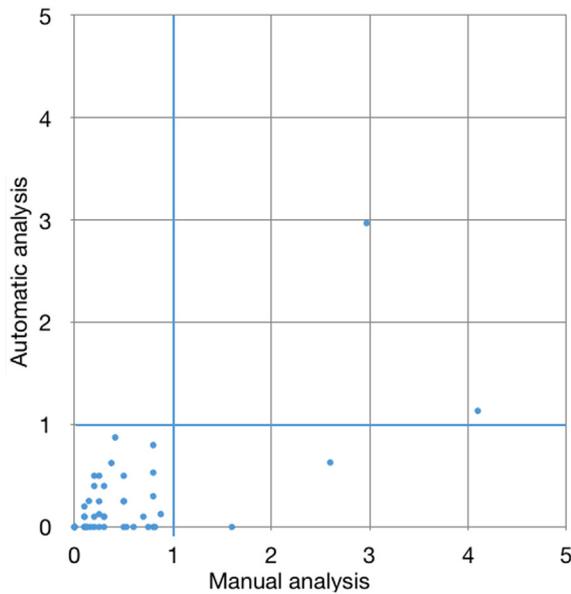


Fig. 3. Central apnea index (CAI) on automatic detection according to manual values. In bold: pathologic CAI threshold = 1.

better, at 67%. In the light of this, sensor placement seems to be a fundamental step which, in our opinion, should be assured by experienced medical or paramedical staff, even in RP at home. Moreover, in Poels et al.'s study, mean age was rather low, at 4.2 years, further suggesting that signal quality is age-related, although admittedly Brockmann et al. had 93% reliability with a median age of 2.8 years [14]. Three studies specifically compared RP at home versus in hospital [3,4,14]. The studies by Alonso-Alvarez and Scalzitti included PSG (gold standard), laboratory RP and home RP. For Alonso-Alvarez et al. [3], apnea and hypopnea detection rates were comparable between the three. Scalzitti et al.'s results [4] were less clear-cut, with better sensitivity for laboratory RP but better specificity at home. Regarding feasibility, Brockmann and Alonso-Alvarez reported comparable reliability for the two RP modalities, whereas Scalzitti found only 67% of technically acceptable recordings at

home, compared to 75% in sleep lab [4]. Home RP is also difficult to associate to transcutaneous or end-tidal CO₂, for reasons of equipment, although this is a very useful signal in children, especially in case of nocturnal hypoventilation as found with underlying neuromuscular or pulmonary pathology (type-3 OSAHS) or associated obesity (type II) [2]. Fig. 4 presents a decision-tree for PSG vs. RP and home vs. hospital, according to age and type of suspected OSAHS.

As respiratory polygraph, we chose the Nox T3 (Nox Medical Inc. Reykjavik, Iceland), validated for SDB exploration in adults [4], for its ergonomics and sensor miniaturization. The finger sensor is connected to a watch fixed on the wrist, which is in turn connected up to the central monitor by Bluetooth[®], reducing the number of cables lying on the child. It should be borne in mind that, in children, any means of securing the nasal and finger sensors, most subject to displacement, is crucial [21]. It is also useful to have several methods of airflow assessment, in case of nasal cannula displacement; extrapolation from respiratory flow assessed by the inductance belts here served as failsafe. Other polygraphs have proved effective in SDB exploration in children [22,23], with complementary signals that are useful in pediatrics, such as substernal recording of airflow and respiratory effort, and respiratory effort assessed by mandibular movement, but requiring more sensors and poorer ergonomics.

Data are sparse regarding automatic analysis of respiratory events by the RP software itself. Khirani et al. found automatic analysis to correlate well with manual analysis in CPAP surveillance [24]. The present study was the first to assess automatic analysis of RP in diagnosing OSAHS in children, and found significant differences with respect to manual analysis, with a clear tendency to overestimate incidence (Table 2). One possible explanation is that the algorithms were derived from those used in adults, for whom RP was initially developed, whereas obstructive respiratory profiles differ greatly between the two populations [21]. Central apnea seemed better assessed than obstructive events: for a pathologic AHI threshold of 1 [1,18], automatic analysis provided 100% specificity, but low sensitivity, at 50%. Practically speaking, child RP interpretation can use automatic analysis of central apnea with manual correction for non-detected events. For obstructive events, the overestimation of apnea and of hypopnea is considerable, and two approaches are possible: manual correction, basically consisting in one-by-one elimination of false positives, or initial elimination of all obstructive events followed by complete manual reassessment. Given the large number of events to be eliminated

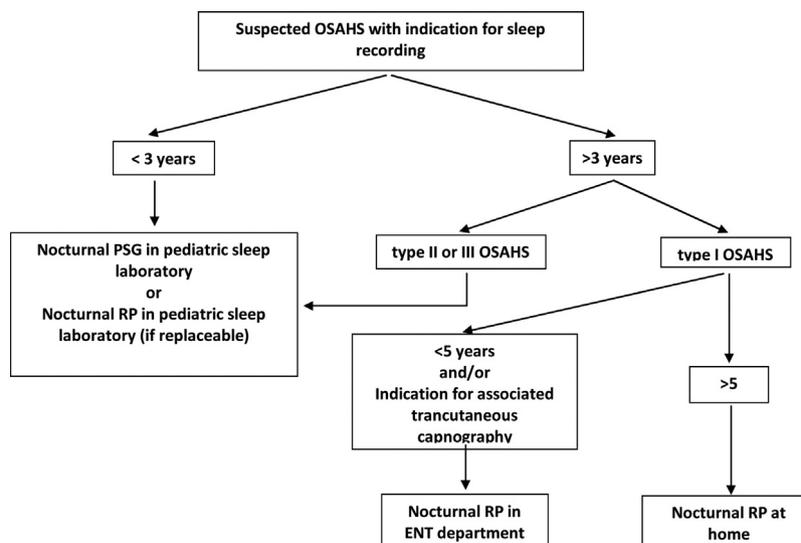


Fig. 4. Choice of type and place of recording according to age and type of suspected OSAHS on Capdevila-Gozal classification [15]. PSG: polysomnography; RP: respiratory polygraphy; OSAHS: obstructive sleep apnea/hypopnea syndrome.

(a mean 102.5 obstructive apneas and hypopneas in the present series), we opted for the second strategy, which is far less time-consuming.

5. Conclusion

RP in children was reliable in routine hospital ENT department practice, with excellent device acceptance and a satisfactory rate of technical acceptability. The children's age influenced overall examination quality, with a particular threshold at 3 years. Implementing RP in a non-specialized hospital setting has the advantage of allowing nursing monitoring of sensor positioning, and the possibility of associating CO₂ measurement, especially in case of suspected type II or III OSAHS [2]. Sensor fitting by a trained physician or nurse seems to be a guarantee of reliability. Over the age of 5 years, in the absence of particular diathesis (suspected type 1 OSAHS), RP may be conducted in the child's home, with possible tele-monitoring, as reported in adults [25]. This associates the advantages of home recording (familiar sleep environment, lower costs, and easy availability) with the possibility of remote real-time signal quality monitoring, enabling the parents to reposition sensors if need be. It is a new technical challenge, worth assessing in children.

Automatic analysis of respiratory events is not reliable in children, except for central apnea: it greatly overestimates the incidence of obstructive apnea and hypopnea, requiring either manual correction of each event, or, as we recommend, eliminating all events from the outset and having them reassessed by a physician trained in pediatric sleep disorder. Automatic detection of central apnea can be contributive, but still requires rigorous manual reassessment, due to the risk of false negatives.

Disclosure of interest

The authors declare that they have no competing interest.

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