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Review Article

Home Respiratory Polygraphy-An Alternative for the Diagnosis of Pediatric Obstructive Sleep Apnea Syndrome

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Abstract

Obstructive sleep apnea syndrome is one of the most common types of sleep-disordered breathing in children and is characterized by partial or complete obstruction of the upper airways during sleep with repeated episodes of airflow cessation, reduction in blood oxygen saturation and sleep disruption to restore patency of the upper airways. Because polysomnography, the gold-standard test for the diagnosis of obstructive sleep apnea, is a costly procedure with technical difficulties, home respiratory polygraphy is used as an alternative diagnostic method. This review seeks to summarize the utility of home respiratory polygraphy in detecting obstructive sleep apnea syndrome and to show if it can be used as a substitute for polysomnography in children.

Keywords: Children; Obstructive sleep apnea; Polysomnography; Respiratory polygraphy

1. Introduction

Obstructive sleep apnea syndrome (OSAS) is a sleep disorder characterized by partial or complete obstruction of the upper airways during sleep with repeated episodes of airflow cessation, reduction in blood oxygen saturation and sleep disruption to restore patency of the upper airways [1]. It is relatively frequent in children [2], affecting 1-4% of the pediatric population [3].

The pathophysiology of OSAS in children is complex and poorly understood. OSAS is characterized by increased upper airway resistance during sleep due to airway narrowing. During daytime, when the patient is awake, the upper airway collapse is prevented by an increased pharyngeal neuromuscular tone [4]. Upper airways obstruction due to adeno-tonsillar hypertrophy is the major risk factor for OSAS in children, and it occurs after repeated upper airways infections [5]. This explains the highest incidence of obstructive sleep apnea between 4 to 6 years of age [1]. Obesity is another important risk factor for OSAS in children due to the increased oro-pharyngeal adipose tissue that will reduce the airways size and will increase airways resistance [6]. Other risk factors for OSAS in children are represented by structural factors related to the skeletal structure of the face (deviated nasal septum, retrognathia or micrognathia, macroglossia, malocclusion, midfacial hypoplasia), neuromuscular factors (hypotonia, cerebral palsy, vocal cord paralysis and muscular dystrophy) [1, 7].

Children with OSAS have breathing difficulties during sleep (mouth-breathing, snoring, increased respiratory effort, apnea), that can lead to behavior problems during daytime (fatigue, mouth-breathing, hyperactive behavior, anxiety, headache, attention deficits and also difficulty concentrating) and development of health related complications.

2. Diagnosis of OSAS

Snoring is a common symptom of upper airway obstruction [1]. It may occur alone (simple snoring) or within OSAS. It is essential to distinguish between simple snoring and snoring associated with OSAS [8]. A person heaving only simple snoring has no other breathing disorders or behavior problems during daytime [1]. The diagnosis of OSAS starts with clinical history and the physical examination. Because, adeno-tonsillar hypertrophy is the most common cause for OSAS in children, upper airway examination is necessary to be performed to assess the size of tonsils and adenoids in relation with oro- and nasopharyngeal space. It is important also to find other disorders that could be responsible for upper airway obstruction (deviated nasal septum, retrognathia or micrognathia, malocclusion, large tongue, high arched) [1, 7]. Medical history and the physical examination are not sufficient to perform the diagnosis of OSAS [9]. Further investigations are necessary.

Diagnosis of pediatric OSAS may be difficult because of the lack of universally accepted criteria for the syndrome and also because children will not always easily tolerate complex recording devices in and around them, and these can represent a problem from the technical point of view [10]. Polysomnography (PSG) is the gold-standard test recommended for the diagnosis of OSAS. It requires to be performed in a sleep laboratory and involves monitoring the following parameters:

- the airflow through the nose and mouth, with an oro-nasal thermistor or a nasal cannula;
- snoring;
- blood pressure and the heart rate;
- blood oxygen level, measured by pulse oxymetry;
- carbon dioxide (CO_2) in exhaled air (end-tidal CO_2), measured by capnography;

- thoracic and abdominal movements;
- electrocardiography (ECG);
- brain electrical activity by electroencephalogram (EEG), being helpful to inform whether the patient is awake or asleep, providing a more accurate assessment of the presence of OSAS and its severity [11];
- electrical activity of muscles and the diaphragmatic effort by electromyogram (EMG);
- eye movement by electrooculogram (EOG).

Nocturnal PSG is indicated when the clinical history and physical examination suggest a possible diagnosis of OSAS [12]. Because it is a costly procedure with technical difficulties, other alternative methods, more simple and less expensive are being tried to assess the presence of OSAS such as audio recording of breath sounds, nocturnal video recording, nocturnal pulse oximetry and respiratory polygraphy [1]. American Academy of Sleep Medicine (AASM) has classified portable monitors into four types according to the American Sleep Disorders Association [13]. They are represented in Table 1 with specifications for each category.

	Type 1 **Full attended PSG**	**Type 2** **Full unattended PSG**	**Type 3** **Limited-channels devices**	**Type 4** **Continous single- or dual bioparameter recording**
Channels	≥ 7	≥ 7	≥ 4	≥ 1
Parameters	EEG EOG Chin EMG ECG Airflow Respiratory effort SpO_2	EEG EOG Chin EMG ECG Airflow Respiratory effort SpO_2	- - - ECG or heart rate Ventilation (≥ 2 channels) • respiratory movements,or • respiratory movements and airflow SpO_2	- - - - Airflow SpO_2
Body position	Documented or objectively measured	May be objectively measured	May be objectively measured	Not measured
Leg movement	EMG or motion sensor (desirable)	EMG or motion sensor (desirable)	Can be recorded	Not recorded
Personnel	Constant attendance	Not in attendance	Not in attendance	Not in attendance

Table 1: Assessment methods for sleep-disordered breathing [13].

Overnight respiratory polygraphy (RP) is a type 3 portable monitor and represents a continuous recording during night of several parameters: nasal airflow (by nasal cannula or an oro-nasal thermistor), thoracic and abdominal movements, heart rate and oxygen saturation. Some RP devices can also register snoring sounds, legs

movements and changes in body position during sleep. It can also be performed at home, having the advantage of leaving the child in its natural sleeping environment, which will improve the comfort and compliance of the patient [1]. Subjects wear the device during all night, and it continuously records the parameters mentioned above. The next day, in the morning, the recording is downloaded and the recorded data can be analyzed automatically using specific software, but also manually.

2.1 Sleep events and indices. Understanding the results

According to the AASM Manual for the Scoring Sleep (2017), apnea in adults is defined as a 90% reduction in airflow for at least 10 seconds [14]. In children, apnea is defined as a 90% reduction in airflow for at least 2 breaths duration, even if they are less than 10 seconds duration [14]. The duration of the apnea is measured from the end of the last normal breath to the beginning of the first breath that achieves the pre-event baseline inspiratory excursion [14]. A specific time in seconds cannot be applicable in children because a normal respiration varies from 12 breaths per minute in an adolescent up to 60 breaths per minute in a newborn.

There are three types of apnea: obstructive, central, and mixed apnea.

The definition of **obstructive apnea** is based on criteria mentioned above plus increased inspiratory effort thorough the entire period of decreased airflow [14]. It occurs when an upper airway obstruction exists. Due to the obstruction, the airflow is reduced, but the thoracic and abdominal respiratory movement are present; usually is accompanied by a reduction in oxygen saturation (SpO_2), followed by an arousal and resume of normal breathing [14].

Central apnea is common during sleep in children [15]. Because is frequently seen in healthy children, is scored only if the event lasts ≥ 20 seconds, and is associated with either arousal or ≥ 3% desaturation [14]. There is no obstruction of the upper airways like in obstructive apnea; the problem is in the connection between the brain and the respiratory muscles that controls the breath. The episode of a central apnea is not accompanied by respiratory effort; so the recording will show an absent airflow with no thoracic and abdominal respiratory movements.

Mixed apnea meets the criteria for apnea in the absence of inspiratory effort in the first part of the event, followed by resumption of inspiratory effort before the end of the apnea [14]. It consists of a central apnea followed by an obstructive component.

Hypopnea is defined as a 30% reduction in airflow; the event lasts at least two missed breath from the end of the last normal breathing amplitude and it is associated with either an arousal or ≥ 3% desaturation [14].

RP is helpful to confirm the diagnosis of OSAS, but also in assessing its severity by calculating the apnea-hypopnea index (AHI), oxygen desaturation index (ODI) and respiratory disturbance index (RDI) [16]. AHI represents the number of apneas and hypopneas per hour of sleep and is used to indicate the severity of OSAS. Unlike PSG, RP does not include EEG, so it cannot differentiate between awake and sleep periods, and therefore the number of apneas and hypopneas must be divided by total recording time instead of sleep time [17]. This results in a systematic underestimation of AHI, so RP tends to underestimate OSAS diagnosis and OSAS severity [11] and, can affect the clinical decision mostly in patients with mild and moderate OSAS [18]. That is why, having a patient with suspected OSAS but

with a negative RP, is necessary to perform PSG for further diagnostic evaluation [11]. Marcus et al. analyzed an overnight PSG of 50 healthy pediatric patients. They find out that the polysomnographic results in the pediatric population are different from those in adults. In children, a patient who has at least one episode of apnea or hypopnea per hour of sleep is considered to have OSAS [19]. According to Tsai, mild OSAS is defined as an AHI ranging from 1 to 4, moderate OSAS as an AHI ranging from 5 to 10 and severe OSAS as an AHI of more than 10 episode of apnea/hypopnea per hour of sleep [16].

RP also has an integrated pulse-oximeter, so it records the oxygen saturation. This allows determining the oxygen desaturation index (ODI), which represents the number of times per estimated sleep duration (time in bed) that the blood's oxygen level drops by ≥ 3% in the absence of moving artifacts [20]. ODI is a good mark for predicting the presence and the severity of OSAS in children [16]. The ODI values can be correlated with AHI [16] and both are used to indicate the severity of OSAS [16, 21].

RDI reports the respiratory events during sleep (apneas and hypopneas), but unlike the AHI, it also includes respiratory-effort related arousals (RERAs) [22]. RERAs is defined as a sequence of breaths ≥ 10 seconds characterized by increasing respiratory effort that does not meet criteria for AASM apnea or hypopnea, but do disrupt sleep [22].

2.2 The utility of home RP for the diagnosis of OSAS

PSG is the gold-standard investigation recommended for diagnosis of OSAS. It is a complicated, time-consuming, expensive procedure [23] and less accessible. Studies have shown that RP can be used as an alternative method to PSG [24-27]. RP is a portable monitoring device, a home recording device, less expensive and more accessible [1].

In adult patients, RP can be used as a substitute for PSG [28]. Candela et al. performed a study on 103 adult patients with suspected OSAS. They tried to validate a cardiorespiratory polygraphy system by comparing it with PSG. The Bland and Altman method was used to assess the agreement between these 2 methods. For an AHI of 10, the best cut-off point determined by manual cardiorespiratory polygraphy analysis was 7.5 (sensitivity 97% and specificity 82%), and for an AHI of 30 or higher, the best cut-off point determined by manual cardiorespiratory polygraphy analysis was 27 (sensitivity 98% and specificity 98%) [29]. They concluded that cardiorespiratory polygraphy system had good agreement with PSG for the measurement of respiratory events [29]. Thurnheer et al. performed a systematic review of 6 studies that compared RP with PSG in adult patients. Patients with suspected OSAS underwent RP and PSG. The pre-test probability for OSAS was 64% [26]. The post-test probability after a positive result was within a range of 98% (positive likelihood ratio of 23.8) to 90% (positive likelihood ratio of 5.7) [26] The conclusion was that in adult patients with suspected OSAS, RP allows an accurate and a simple diagnosis of OSAS [26]. Other studies [24, 25, 28] found a significant correlation of AHI between PSG and a portable sleep monitor device in adult patients.

Few studies have investigated the utility of RP in detecting sleep-disordered breathing in children. All of them were carried out with a small number of patients. Zucconi et al. included in a study 12 children aged 3-6 years with suspected OSAS who underwent PSG and RP. They reported that the specificity of RP was low for RDI>5 and increased only at RDI>10 (71% specificity),

but decreased at 57% with revised RDI, although the sensitivity of RP was satisfactory (80%) [30]. They concluded that home RP is more useful for screening patients with highly suspected moderate-to-severe OSAS [30]. Hammoudi et al. performed a study based on 20 children with suspected OSAS. The aim was to assess the agreement between RP and PSG in children. At the end of the study, there was some agreement between RP and PSG of measuring AHI [31], but they concluded that further studies need to be performed with a higher number of children before making recommendations for routine use of RP in children with suspected OSAS [31].

Studies performed by Brockmann et al. (101 children, median age 2.8 years) [32] and Rosen et al. (850 children, aged 8-11 years old) [33] in patients with sleep- disordered breathing who underwent RP, showed a good positive predictive value of RP with a sensitivity of 88% and a specificity of 98% for an AHI > 5/h [34].

In a study, Scalzitti et al. investigated the ability of a portable monitor to diagnose OSAS in children aged 2-17 years old. An analysis of the sensitivity and specificity of the portable monitor for the diagnosis of OSA compared to the PSG was performed [35]. The sensitivity of home device was best when the device was worn in a sleep laboratory (81.5%), with a specificity of 60%, while the sensitivity was lower (70%), and the specificity only 42.9%, when the monitor was worn at home [35]. The results of the study are probably influenced by the age of the patients, because children under 5 years old had a significantly higher error with the portable device compared to the PSG [35]. The AHI measurements did not statistically differ on the home sleep test from the PSG in children age 6 and older [35].

2.3 Convenience and acceptability

PSG is a complicated procedure. The patient needs to sleep overnight in a sleep laboratory under observation. The technical equipment used to perform PSG in children is the same used for adults [1]. In pediatric patients, an overnight PSG should be performed in a child-friendly environment, heaving one parent beside the child. It is recommended that the parent should sleep in another bed, because its movements can be confused as coming from the patient and may influence the diagnostic procedure [1]. Home RP has the advantage of letting the child in its natural sleeping environment, improving the comfort and the compliance of the patient [1, 13]. No reports have been published regarding the morbidity or mortality associated with RP [13]. In their study, Goodwin et al. reported some unpleasant things that appeared during the night recording: 40% of parents declared that their child slept worse than usual due to the discomfort caused by the nasal cannula [36]. In his study, Poels et al. also reported that 58% of parents complained that the nasal cannula caused irritation [8].

2.4 Diagnostic limitations

The limitations of RP should always be considered. In pediatric patients, one of the technical challenges is to obtain adequate nasal airflow signal [37]. Some children with sleep-disordered breathing are mouth breathers so, in this situation, the nasal airflow signal is reduced [37]. Smaller children have difficulties in accepting the nasal cannula, so that will influence the results of RP. Also, the nasal cannula can be removed during sleep due to the child's movements. In a study, Gudnadottir et al. tried to evaluate the quality of home RP in 60 children with symptoms of sleep-disordered breathing [37]. They found that the majority of home RP performed in the study were unsuccessful and this was due to the loss of the nasal flow signal [37]. The 2015 AASM guidelines, recommends the use of an oro-nasal thermal flow sensor

for scoring apneas and a nasal transducer for scoring hypopneas for PSG [38]. In a study made in 201 children, aged 5-12 years, Marcus et al. found that the success rate of the nasal airflow signal with an oro-nasal thermistor was 92% compared with a 67% success rate when a nasal cannula was used [39]. In their study, Scalzitti et al. found that 89% (32 of 36 recordings) of home RP were technically acceptable, and 67% (24 of 36 recordings) were successful recordings [35]. Their successful results were probably determined by using a combination of nasal cannula and an oro-nasal thermistor. Besides PSG that records EEG, RP cannot identify sleep stages, so arousals or sleep disruptions, are hard to recognize [13]. PSG can also measure the carbon dioxide (CO_2) in exhaled air (end-tidal CO_2), by capnography, used as an indicator of airflow obstruction during sleep [40]. Unlike PSG, RP doesn't measure the CO_2, so cannot distinguish if a low SpO_2 is related to hypoventilation [26].

2.5 Feasibility

A few studies tried to evaluate the feasibility of RP for the diagnosis of OSAS. In 1995, Jacob et al. performed a study on 21 children aged 2-12 years old with adenotonsillar hypertrophy and suspicion of OSAS. At the end of the study, he reported an 83% successful recording rate and he concluded that RP could be used in the routine evaluation of OSAS in patients with adenotonsillar hypertrophy [41].

Goodwin et al. included in a study children aged 5-12 years old. They performed an unattended home RP, and they reported a 91% successful recording rate [36]. Poels et al. also wanted to evaluate the feasibility of using a home cardiorespiratory recording device in 24 children aged 2-7 years old that were scheduled for adenotonsillectomy. From the 24 recording, only seven were successful (29%) [8]. He tried to find some explanations for the differences between his study's results and the other two study's results. One explanation was that Goodwin included in the study older children (aged 5-12 years) with which it is probably easier to collaborate. Also, Poles et al. confirmed that in their study the results of technically acceptable recordings were better in older children (the mean age of the participant with a successful recording was 5.7 years versus 3.6 years for unsuccessful recordings) [8]. Another explanation can be the duration for overnight recording. In adult patients, the minimum recording duration is 360 minutes, but for children no such criteria are established [8]. Poels established for his study a minimum duration of 390 minutes as the criteria for a technically acceptable recording. In contrast, Goodwin considered that a period of 240 minutes is sufficient for a successful technique. However, more studies are necessary to establish the optimal duration of recording. The third explanation for better results in Jacob et al. and Goodwin et al. studies is that the home monitoring devices were set up by a qualified technician [8]. In contrast, in Poels's et al. study, the parents were trained to set up the equipment, and this probably decreased the successful rate [8]. Blanc et al. included in a study 50 children with suspected OSAS, aged 2-11 years old, who underwent RP. The device was well accepted in 98% of cases with the average signal quality 70.8% (86% in children >3 years old, and 25% in children <3 years old) [42].

2.6 Estimation of costs

PSG is a costly procedure with technical difficulties, time-consuming, and also less accessible [23]. Instead, RP is more accessible, with simple sensor setup and with lower cost [42]. Masa et al. tried to evaluate the diagnostic efficacy and the cost of home RP compared with PSG [43]. They reported that the cost of home RP was six times lower than PSG when patients transported

the device and four times lower when data were transmitted by telematics [43]. Quiroga et al. also compared the effectiveness and the costs of home RP versus PSG for diagnosis of OSAS. Compared with PSG, home RP had similar effectiveness but the cost was 421 € lower [44].

3. Conclusions

Snoring is a common symptom of upper airway obstruction. It can occur alone (simple/primary snoring) or as an indicator of OSAS. OSAS is characterized by repeated episodes of hypopnea and apnea during sleep because of the partial or complete obstruction of the upper airways. In children, the most frequent cause of upper airways obstructions is adenoid and/or tonsil hypertrophy. Children with OSAS have breathing difficulties during sleep (mouth-breathing, snoring, increased respiratory effort, apnea) and behavior problems during daytime (fatigue, mouth-breathing, hyperactive behavior, anxiety, headache, attention deficits and also difficulty concentration).

It is essential to distinguish between simple snoring (without episodes of apnea) and snoring associated with OSAS. Clinical history and physical examination are not sufficient to make the difference, so it is important to perform some specific tests. PSG is the gold-standard for the diagnosis of sleep-disordered breathing problems. Because of the technical difficulties and high cost of PSG, other portable monitor devices are available. RP is a type 3 recording device, less expensive, more accessible, and more convenient for the patient, having the advantage to be performed at home.

Few studies have investigated the role of RP for the diagnosis of OSAS in children, by comparing it with PSG, and all included a small number of patients. Some of them showed that RP could be used to diagnose and to assess the severity of OSAS. Despite all the studies performed till now, more research is required to assess the utility of RP for the diagnosis of OSAS in children.

Disclosure of Interest

The authors have no conflicts of interest to report.

Authors' Contributions

SCM and CNȚ(S) conceptualized the study, performed data collection and analysis data and wrote up the various drafts for publication. VS and GAF assisted in the review of the various drafts for publication. All authors have read and approved the final version of this manuscript.

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