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Current hypopnea scoring criteria underscore pediatric sleep disordered breathing

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ABSTRACT

Objective: This is a retrospective study comparing 2007 American Academy of Sleep Medicine (AASM) pediatric scoring criteria and Stanford scoring criteria of pediatric polysomnograms to characterize the impact different scoring systems have upon the diagnosis of sleep disordered breathing in children.

Methods: The diagnostic and post-treatment nocturnal polysomnograms (PSGs) of children (age 2–18 years) consecutively referred to an academic sleep clinic for evaluation of suspected sleep disordered breathing (SDB) for 1 year were independently analyzed by a single researcher using AASM and Stanford scoring criteria in a blinded fashion.

Results: A total of 209 (83 girls) children with suspected SDB underwent clinical evaluation and diagnostic PSG. Analysis of the diagnostic PSGs using the Stanford and AASM criteria classified 207 and 39 studies as abnormal, respectively. The AASM scoring criteria classified 19% of subjects as having obstructive sleep apnea (OSA) while the Stanford criteria diagnosed 99% of the subjects with OSA who were referred for evaluation of suspected sleep disordered breathing. There was a positive correlation between SDB-related clinical symptoms and anatomic risk factors for SDB. Scatter-plot analyses showed that the AASM apnea hypopnea index (AHI) was not only significantly lower compared to the Stanford AHI but also skewed in distribution.

Results: Ninety-nine children were restudied with PSG (9 were initially diagnosed with SDB with AASM criteria, whereas all 99 were diagnosed with SDB with Stanford criteria). All 99 children had been treated and had a post-treatment clinical evaluation and post-treatment PSG during the study period. All 99 children evaluated after treatment showed improvement in clinical presentation, Stanford AHI, and oxygen saturation during sleep.

Conclusion: The AASM scoring criteria classified 19% of subjects as having OSA while the Stanford criteria diagnosed 99% of the subjects with OSA who were referred for evaluation of suspected sleep disordered breathing. The primary factor differentiating the AASM and Stanford criteria was the scoring of hypopneas. The AASM definition of hypopnea may be detrimental to the recognition of SDB in children.

1. Introduction

Contemporary diagnosis and treatment of obstructive sleep apnea–hypopnea syndrome in children rely heavily upon objective evaluation by nocturnal polysomnography. The diverse constellation of symptoms in children with OSA makes it difficult to accurately identify children at risk for OSA. Diagnostic polysomnograms (PSG), therefore, are important in identifying and measuring response to treatment in children with OSA. The apnea–hypopnea index (AHI), a measurement used to summarize the number of abnormal breathing events per hour of sleep, is indisputably the key computed metric of the PSG. The AHI is the \textit{lingua franca} of the sleep world – the basis for diagnosis, prognosis, treatment, and measurement of successful treatment outcomes; yet, its exact definition is controversial.

Since the definition of OSA in children in 1976 [1], studies have shown that children may have clinical symptoms that are related to abnormal breathing patterns during sleep [2,3] but without the presence of apneas or hypopneas meeting established criteria for SDB. Disagreement regarding the definition of an hypopnea, the duration of an electroencephalogram (EEG) arousal, the degree of airflow reduction, and the percentage of oxygen desaturation has been reported [4–8]. The use of different scoring criteria has led to significant variability in the AHI [6–10], subsequently affecting the interpretation of disease severity, the treatment plan for individual patients, and the estimation of prevalence and morbidity for epidemiological purposes. The 2007 AASM scoring criteria [11], emphasizing an old rule for scoring “hypopneas” and adding the “respiratory event related arousal” component without any well-defined criteria for scoring these events, have not resolved this issue.
2. Methods

2.1. The Stanford hypopnea definition and related scoring considerations

The definition of abnormal respiratory events for children was established in the late 1970s and early 1980s with the use of esophageal manometry (Pes) [12–16]. The initial criteria were established by monitoring normal subjects recruited from the general population [3,12–16]. Based on the esophageal pressure monitoring, several patterns have been described as abnormal breathing patterns without complete apneas (i.e., hypopneas) [3,10,12–16]. With Pes monitoring, hypopneas, indicated by increased or decreased (see Fig. 1) respiratory efforts lasting at least three breaths followed by a return to baseline of the Pes signal, were associated with a change in the EEG signal. The EEG changes seen with return to baseline of the Pes signal with non-rapid eye movement (NREM) sleep hypopneas were associated not necessarily with a burst of alpha or alpha/beta frequencies lasting three or more seconds but rather with a delta burst often followed by a very short-lived (between 0.5 and 2 s), fast (alpha, or mixed alpha–beta range) frequency component [17,18]. This is very similar to the pattern defined as “phase A2” when using cyclic-alternating-pattern-CAP-scoring [19] (see Figs. 2 and 3). With the development of the nasal cannula pressure transducer, Stanford Sleep Disorders Clinic now performs recordings without Pes monitoring in about 65% of children, particularly in cases where suspicion for SDB is high at the time of clinical evaluation [17]. In order to adapt the Stanford Pediatric Hypopnea Criteria to studies performed without Pes, we verified the validity of the Pes-derived hypopnea rules when applied to studies without Pes monitoring. To accomplish this, one trained scorer was given records in which nasal cannula pressure transducer, oral thermistor, Pes, intercostal-diaphragmatic and expiratory EMG signals, and inductive plethysmography belts were all simultaneously used to monitor respiration in addition to other standard recommended PSG leads. The scorer reviewed the data in a blind fashion, with and without the availability of the Pes signal in the montage, and analyzed these signals. The Stanford rules for scoring NREM and REM hypopneas without Pes were derived from this work [3,10,17,18]. The definition of “REM sleep related hypopneas” was more difficult without Pes signal because the nasal cannula evaluates changes in flow but not changes in effort (see Fig. 1). The decision was to define them as a decrease in the nasal cannula–pressure transducer flow curve by 20% or more during long bursts (longer than 3 s) of rapid-eye-movements [3] (see Fig. 1). These REM related hypopneas were not associated with a simultaneous increase in chin EMG and burst of fast EEG (alpha/beta frequencies) of three or more.

We also continued to score stages 3 and 4 sleep separately as described in the Rechtsaffen and Kales manual [20], as stage 4 may not be attained or may be curtailed with SDB. The presence of only stage 3 NREM sleep may indicate a potential health problem in young individuals [21], as it is often a pattern with repetitive phase A2 CAP sequences [19]. We used the AASM rules as published [11], and the Stanford rules are indicated in Tables 1 and 2 (see also Figs. 1–3).

2.2. Protocol

In the sleep center, all polysomnograms were scored initially by technologists using the “Stanford scoring criteria,” (with inter-scorer agreement of 92.4%) subsequently reviewed by the same experienced sleep specialist; clinical recommendations were made to parents and referring physicians according to clinical practice.

In part one of this study, the diagnostic PSGs were rendered anonymous and rescored by one individual who was blinded to patient history and clinical recordings. The records were scored using standard AASM pediatric respiratory rules and then rescored using the Stanford pediatric respiratory rules. Slow wave sleep was also rescored with both stages 3 and 4 NREM rather than just stage N3 NREM. Anonymous clinical data were then matched with rescored anonymous PSGs.

In part 2 of this study all children treated for their SDB who underwent post-treatment PSG during the study had PSG and

Fig. 1. Example of several REM sleep Stanford hypopnea determined with Pes recording. Nasal cannula recording is on channel 13 from the top and Pes recording is just below (channel 14-red). Per convention inspiration is up for nasal cannula and down for Pes. One can see the decrease in inspiratory efforts on the Pes recording associated with phasic REM and the associated decrease in the amplitude of the nasal cannula curve involving the same breaths, indicative of an “hypopnea” during phasic REM sleep. The finger plethysmography curve (channel 9-) shows two different types of fluctuations: some related to the phasic REM in association with burst of rapid-eye-movement, but also changes associated with return to normal breathing as can be seen on number 1 of the sequence of Stanford hypopneas (increase in sympathetic activity is indicated by a downward movement of the signal). As can also be seen on that number 1 hypopnea, this return to normal amplitude of nasal cannula curve is associated with a change in the EEG (channels 1–4) with a burst of high amplitude waves. There is no change in EMG, the oxygen saturation change in the number 1 hypopnea is 1%, the maximum oxygen saturation change in the total sequence is 2%. Note that when monitoring intercostal-diaphragmatic EMG, channels 18 and 19, one can also perceive the change in efforts, it is better seen when performing an “integration” of the signal, as reported by Strohs et al. [45] on channel 17. The abdominal band in this case gives also information, but the raw signal on the abdominal band may not be always as clear, depending on position. (For interpretation of the references to color in this figure legend, the reader is referred to the web version of this article.)
clinical history rendered anonymous. PSG were scored twice following the same protocol by the same individual blinded to patient clinical evaluation and results. Once PSGs had been scored, anonymous clinical data were again matched with the scored PSGs. The study was approved by the Stanford University Institutional Review Board.

2.3. Subjects

The patients, aged 2–18 years, involved in this retrospective analysis underwent diagnostic PSGs at the Stanford Sleep Disorder Clinic from August 2009 to June 2010.

The Stanford Sleep Disorders Clinic is a tertiary referral center. The majority of children were referred to the clinic by their pediatrician but 15% of the patients were self-referrals. Information including current medications and previous treatment history were obtained from referral documents and during the clinical interview. Children with neuromuscular disorders, genetic disorders (e.g., Downs syndrome, Prader-Willi syndrome), endocrine syndromes, major craniofacial dysmorphia (Pierre-Robin, Crouzon, etc.), or other chronic medical disorders that could be associated with SDB were excluded. Children with previously unsuccessful OSA treatment were also excluded from the study as well as overweight/obese subjects (body mass index [BMI] calculated based on standardized percentile curves [22]).

2.4. Clinical evaluation

2.4.1. Clinical symptoms and history

During clinic visits all children had pediatric evaluations including BMI generated from weight and height [22], vital signs, and neck circumference measurements. The Pediatric Sleep Questionnaire was completed [23], and thorough sleep/wake and sleep medicine histories were obtained in addition to medical, surgical, familial, social histories, and review of systems.

2.4.2. Anatomic examinations for airway

All patients underwent a systematic airway examination using subjective scales. The examination followed a standardized approach. Anatomic structures including nasal passage, oral cavity, and craniofacial structures were examined. The anatomical evaluation was previously published and involved a thorough nasal evaluation [24]; the intra-oral examination included hard palate, soft palate, tongue, buccal mucosa, dental occlusion, and appreciation of high and narrow palatal vault and/or posterior cross bite [25]. The relationship between the soft palate and tongue was defined by the Mallampatti scale [26]. The 4-point Friedman scale from 1+ to 4+ was used to evaluate the horizontal dimension of tonsils [27]. Finally, evaluation of cranial-facial structures was performed [28], with class II malocclusion, overjet greater than 2 mm, dental crowding, and impacted lower molar teeth identified as a sign of a retrognathic mandible and [29] characteristics such as long faces and retrognathic upper and lower jaws as positive anatomic predisposing factors for SDB.

2.4.3. Clinical impression

The clinical impression of pediatric sleep-disordered-breathing was formed by two or more physicians after independent examinations, based on clinical symptoms and physical examination findings.

2.4.4. Polysomnography (PSGs)

All children underwent an overnight in-laboratory polysomnography with lights out guided by usual home bedtime and wake.
time routine. The polysomnographic variables monitored during nocturnal sleep are outlined in Table 2. Esophageal pressure (Pes) was monitored as an additional parameter in a small number of subjects, but this channel was masked during this investigation. Instrumental and biological calibrations were performed before sleep onset. Continuous video monitoring was conducted during total recording time. The minimum recording time was 7 h. Typically, one parent or guardian was present in the room during the polysomnographic study.

2.4.4.1. Principles of management and follow-up. As mentioned above, patients and parents/guardians and referring physicians were informed with the PSG results at follow-up in the clinic and/or by formal letters. At the sleep clinic, for patients with a
Table 2
Stanford Hypopnea definition.

(A) Montage requested
EEGs (4 derivations) Chin and leg EMGs, two EOGs EEG (modified D2 derivation) body position sensor; respiratory montage: nasal cannula pressure transducer, mouth thermistor, thoracic and abdominal inductive plethysmography belt, neck microphone, finger pulse oximetry giving oxygen saturation curve and finger plethysmography curve, intercostals-diaphragmatic and abdominal expiratory muscle EMG, continuous video-monitoring. Transcutaneous CO2 is recommended and can be replaced by end-tidal CO2. Pes should be monitored if presence of abnormal breathing during sleep is questioned and if clinical presentation is uncertain. Bio-calibration with documentation of inspiration and expiration with spontaneous breathing and on command is present on tracing.

(B) Definition (without usage of Pes)
An hypopnea is a breathing events lasting at least longer than two breaths (i.e., 3 breaths) independent of age of the child (1–18 years). It is scored based on nasal cannula pressure transducer; (scoring without esophageal manometry) it is associated with a decrease of the curve by 20% compared to the 3 min prior baseline recording. An hypopnea begins with the drop of the nasal cannula curve to reach a 20% drop during one breath. The hypopneas ends when the nasal cannula returns to baseline. The duration of the hypopnea is calculated from the inspiratory movement of the 1st abnormal breath till the inspiratory movement of the first normal breath. This first breath may show indication of increased movement amplitude above prior baseline volume and associated with short-lived hyper-ventilation.

Investigation of other respiratory signals should be perform for the breaths involved in the hypopneas:
1. Checking presence/absence of increase in inspiratory muscle EMG simultaneously with movement and change in amplitude of the inductive thoracic and/or abdominal belts; such association indicates presence of an obstructive hypopneas. If there is a decrease in all of the above signals during the hypopneas, a “central hypopneas” as seen in association with phasic events of REM sleep may be suspected but cannot be affirmed without Pes recording.
2. Checking presence or absence of mouth breathing based on oral signal.
3. Checking presence of simultaneous inspiratory abdominal EMG discharge. If snoring is present note presence of progressive increase in discharge of signal and temporal relationship with inspiratory or expiratory EMG discharges.

EEG association
(a) End of hypopneas can be temporally associated with a burst of delta waves lasting at least 2 s with or without presence of a short-live (one half to two seconds) low amplitude fast frequency pattern between (8 and more Hz and not responding to spindle definition), with a short low amplitude fast frequency pattern visually discernable (1 s), or with an AASM defined EEG arousal.
(b) The EEG pattern can be associated with a change of the plethysmographic curve with a visually recognizable descending and short lived curve pattern indicative of a sympathetic activation.

With usage of Pes
Hypopneas are still defined based on nasal cannula, but Pes helps in recognition of hypopnea-onset with change in Pes amplitude compared to prior recorded breaths, and allows to quantify (after Pes calibration) the amount of change in inspiratory effort associated with each breath. Patterns such as “Pes crescendos”, “sustain continuous effort”, and “Pes reversal” are systematically search (??) for and will define increase inspiratory efforts as published. Pes also allows to define an hypopnea with a decrease effort (such as seen in REM sleep see Fig. 1).

Stanford AHI > 1.5 events/h, treatment recommendations were given according to the clinical symptoms, clinical findings including anatomic risk factors, PSG results based on Stanford AHI, and desire of patients and referring physicians. Positive airway pressure (PAP) titration and treatment were arranged as necessary. Whenever anatomic predisposing factors were noted, a consultation with a senior sleep surgeon and an experienced orthodontist was scheduled if requested.

Surgical referral to address specific anatomic problems such as enlarged turbinates (radiofrequency treatment) and enlarged adenoids and tonsils (tonsillectomy and adenoidectomy) was considered when appropriate. Rapid maxillary expansion was recommended for patients with high and narrow palatal vault and narrow nasal passages [29]. Skeletal surgery such as correction of septum deviation and maxillofacial surgery for retrognathia were mentioned if age- and patient-appropriate. If allergies/atopy, particularly nasal allergies, were present, the patient was also referred to a pediatric allergist. Follow-up post treatment polysomnograms were recommended to all patients.

When PAP treatment was prescribed, patients were scheduled to return for follow-up visits 3–4 weeks after initiation of treatment. Adjustment of PAP settings was performed as indicated, and post-PAP PSG recordings were performed after establishment of good compliance. For patients who underwent surgery or maxillary expansion, a follow-up PSG was requested 3–6 months after the intervention to allow time for soft tissue adjustment after surgery and/or maxillary expansion.

Independent of treatment selection, all patients or family members filled out the same validated questionnaires and underwent the same standardized evaluation post-treatment as at entry.

2.5. Research study scoring

For the purpose of this study, unmarked PSGs were blindly re-scored twice by one individual using AASM and Stanford scoring criteria.

2.5.1. Wake and sleep stages
In this study wake and sleep stages were scored according to the international manual and criteria [11,20,30,31]. In order to appreciate the importance of slow wave sleep [21] using the Stanford criteria, stage 4 NREM sleep-stage scoring was systematically separated from total slow wave sleep using Rechtschaffen and Kales criteria [20]. EEG arousal (≥ 3 s) [11,30,31] was scored independently from respiratory events.

2.5.2. Oxygen desaturation
Oxygen desaturation was checked to verify if artifact (identified as deformity of finger plethysmography waveform), coexisting with sympathetic related peripheral hypo-perfusion as indicated by the plethysmography curve, was present and was marked accordingly [32]. After removing artifact by manually checking the plethysmography waveform, a desaturation module was run to label all desaturation events according to preset criteria (≥ 3%).

2.5.3. Respiratory events
Abnormal breathing patterns were identified and labeled as defined using AASM criteria [11] and those outlined in Tables 1 and 2 for the Stanford criteria. The scoring of respiratory events was based on total sleep time and was used for calculation of the apnea–hypopnea index (AHI). The designation of an abnormal AHI was defined according to the International Classification of Sleep Disorders as greater than one event per hour [33] for AASM scoring and greater than 1.5 events per hour for Stanford scoring [17].

Tabulated data were extracted after blinded scoring of the PSGs; these data included the subject study research ID number, de-identified information including age, sex, weight, height, and calculated BMI, pulse rate, respiratory rate, blood pressure, Stanford AHI, AASM AHI, oxygen saturation nadir, total recording time, sleep period, sleep time, sleep efficiency, sleep onset, stage 1 shift, stage shift, number of awakenings, number of REMs, REM-onset, REM latency, percentage, and onset of each sleep stage, index of apnea and hypopnea of different characteristics (obstructive or central;
non-REM or REM; supine or non-supine), oxygen saturation (mean, minimal, and maximal; in awake, non-REM, REM, and total sleep time), and periodic leg-movement (PLM) index.

2.6. Clinical data analysis

For the purpose of the study the following data were summarized.

2.6.1. Clinical Information

Collection of the PSG and use of a standardized spreadsheet for clinical complaints, history, physical examination, and demographic information were used by physicians in the sleep clinic to standardize clinical data collection and were provided as de-identified data.

Subjects were grouped by ages 2–5, 6–11, and 12–18. Sleep-related symptoms were subdivided as present at bedtime, occurring during sleep (either wake after sleep-onset or wake-up time), and daytime symptoms. For bedtime symptoms, routine hypnotics use or antihistamines-as-hypnotics use at bedtime were considered to be signs of insomnia. Daytime symptoms included problems paying attention in class, learning difficulties, hyperactivity, headache, daytime fatigue, daytime sleepiness, naps, unrefreshing nap, and developmental delay. For each category, the total number of symptoms was calculated without weighing specific items. The prevalence of each symptom was computed.

The results of the each anatomic evaluation was presented as a sum of predisposing factors for SDB that had been obtained using mostly subjective scales even if standardized in the clinic. We also developed a simple scoring system based on presence of location of anatomic risk-factors at several levels: “1” for 1 level of obstruction, “2” for 2 levels of obstruction, and “3” for 3 levels of obstruction (score of 3 indicated presence of nasal, pharyngeal and mandibular-hypopharyngeal risk-factors). The maximum possible score was 3.

Results of the sleep and respiratory PSG data were added to the clinical information for analysis.

2.7. Statistics

Descriptive analysis was performed for the summary statistics. Due to non-normal and skewed distribution noted for both Stanford AHI and AASM AHI, even after logistic transformation, inferential analysis was performed using Wilcoxon rank test, Mann–Whitney test and Spearman correlation. Wilcoxon rank test was used to test data obtained after and before treatment. Mann–Whitney test was used to compare data obtained between different age and gender groups. Spearman correlation was used to test correlations between Stanford AHI, AASM AHI, clinical symptoms, and anatomic predisposing factors.

3. Results

3.1. Patient characteristics

A total of 209 (83 girls) patients over the designated 1-year period met inclusion and exclusion criteria. There were 51 children aged 2–5 years, 75 children aged 6–11 years, and 83 preteens/teens aged 12–18 years. Demographics are presented in Table 3. None of the subjects were overweight according to standardized percentile curves for body-mass-index. Prior to coming to the Stanford Sleep Disorders Clinic, 43 children were previously seen in other sleep clinics and 135 patients (73 [34.9%] aged 2–5 years; 23 [30.7%] aged 6–11 years, and 39 [47%] aged 12–18 years) were receiving medications for their clinical symptoms. These medications are listed in Table 4; medication was either started or increased in 36 of the 43 children (83.7%) in whom diagnosis of OSA was ruled out in a prior sleep evaluation based on AASM scoring.

Prevalence of clinical symptoms in each group is summarized in Table 5. The number of complaints from individual patients ranged from 1 to 26 (7.5 on average). Anatomical predisposing factors are presented in Table 6; all subjects had anatomical risk factors. For individual subjects, total anatomical factors identified in one individual ranged from 3 to 19 (9.3 in average). The simple scoring method evaluating presence of anatomical risk factors at different upper airway levels shows that 166 subjects (79.4%) had risk factors at three levels.

3.2. PSG results by two scoring systems

Comparison between Stanford AHI and AASM AHI is shown in Table 7.

As shown in Fig. 4 (scatter-plot) the majority of children had an AASM score of 0 independently of age group. Comparison of scoring of apnea and hypopneas separately showed that nearly all events scored as “apnea” using the AASM scoring system were also scored as “apneas” using the Stanford scoring system. Only four events in the total analysis were scored differently: two events scored as “apnea” using the Stanford system were scored as “hypopneas” using the AASM scoring system, and two events scored as “hypopneas” with the Stanford system were scored as apnea with AASM scoring. However, all AASM events were scored using the
Stanford scoring system either as apnea or hypopneas. The discrepancy between the two scoring methods was due to differences in hypopnea scoring, which were scored much more frequently with the Stanford system. Using Rechtschaffen and Kales sleep staging criteria, there were 33 (15.8%) children (8 [15.7%] aged 6–11 years, and 10 [12.0%] aged 12–18 years) who reached the post-treatment time requested before re-evaluation, influencing PAP titration, surgery, or maxillary expansion, or had not completed orthodontic rapid maxillary expansion. Their demographics and initial treatment results using AASM criteria (AASM-AHI >1), whereas all 99 were considered to be abnormal using Stanford AHI criteria (Stanford AHI >1.5). Those without follow-up studies were awaiting PAP titration, surgery, or maxillary expansion, or had not reached the post-treatment time requested before re-evaluation, correlated to each other and to anatomic predisposing factors. However, there was no significant correlation between PSG score and either AASM or Stanford score, and between PSG score and either clinical symptoms or anatomical factors (Table 8). In groups aged 2–5 and 12–18 years, PSG scores inversely correlated with minimum oxygen saturation (i.e., the lower the score, the greater the risk of lower oxygen saturation), but this correlation was not significant in the group aged 6–11 years.

3.4. Treatment recommendations and post-treatment follow-up

Positive airway pressure (PAP) treatment was considered for 107 patients (52%) as a first-line treatment; the other patients (n = 100) were directed to otolaryngological treatment with or without orthodontic treatments. Social factors, such as decision not to have surgery before end of school-year, influenced initial treatment selection.

During the survey period, 99 patients (47.4%) were restudied by PSG post-treatment interventions and none had completed orthodontic rapid maxillary expansion. Their demographics and initial information at entry are presented in Table 3. Of these 99 clinic patients with pre- and post-treatment PSGs, nine had abnormal pre-treatment results using AASM criteria (AASM-AHI >1), whereas all 99 were considered to be abnormal using Stanford AHI criteria (Stanford AHI >1.5). Those without follow-up studies were awaiting PAP titration, surgery, or maxillary expansion, or had not reached the post-treatment time requested before re-evaluation.

### Table 5

<table>
<thead>
<tr>
<th>Associated with sleep</th>
<th>2–5</th>
<th>6–11</th>
<th>12–18</th>
<th>Total</th>
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</thead>
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<tr>
<td>Event/h</td>
<td>51(68.0)</td>
<td>73(31.0)</td>
<td>12(9.4)</td>
<td>166(100.0)</td>
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<tr>
<td>Median</td>
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<td>11.0</td>
<td>15.8</td>
<td>14.8</td>
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<tr>
<td>Quartile range</td>
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<td>(8.2, 15.9)</td>
<td>(10.4, 21.2)</td>
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</table>

### Table 6

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<th>6–11</th>
<th>12–18</th>
<th>Total</th>
</tr>
</thead>
<tbody>
<tr>
<td>Event/h</td>
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<td>78(94.0)</td>
<td>78(94.0)</td>
<td>225(100)</td>
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<td>Median</td>
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<td>15.8</td>
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<tr>
<td>Quartile range</td>
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<td>(15.8, 24.0)</td>
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### Table 7

<table>
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<th>Clinical symptoms/signs</th>
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<th>12–18</th>
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<td>Number (percentage)</td>
<td>51(68.0)</td>
<td>73(31.0)</td>
<td>12(9.4)</td>
<td>166(100.0)</td>
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<tr>
<td>Median</td>
<td>14.8</td>
<td>11.0</td>
<td>15.8</td>
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<tr>
<td>Quartile range</td>
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<td>(8.2, 15.9)</td>
<td>(10.4, 21.2)</td>
<td>(10.4, 21.2)</td>
</tr>
</tbody>
</table>
or had not undergone treatment yet, as subjects involved in the review were collected over time.

3.5. Post treatment results

Forty-five patients completed PAP titration. The oxygen saturation nadir improved from 89.5 ± 4.6 to 91.8 ± 4.3 percent (p = 0.001). Stanford AHI was significantly reduced from 18.6 ± 8.6 to 2.2 ± 1.5 (p = 0.001). Stanford AHI was significantly reduced from 18.6 ± 8.6 to 2.2 ± 1.5 (p = 0.001).

Forty-four other patients returned for follow-up PSG 3–6 months after completion of tonsillectomy and adenoidectomy with/without radiofrequency treatment of turbinates. Overall, Stanford AHI was significantly reduced from 15.5 ± 9.5 to 11.54 ± 7.3 events/h (p = 0.033). Minimum oxygen saturation was significantly improved from 90.2 ± 3.9% to 91.7 ± 2.7% (p = 0.001).

Table 8
Significant Correlation between Stanford AHI and AASM AHI, and between clinical-symptoms and anatomic factors.

<table>
<thead>
<tr>
<th>Age groups</th>
<th>Stanford AHI and AASM AHI</th>
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<tr>
<td>2–5</td>
<td>0.461**</td>
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<tr>
<td>6–11</td>
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<tr>
<td>12–18</td>
<td>0.324**</td>
</tr>
<tr>
<td>DS</td>
<td></td>
</tr>
<tr>
<td>2–5</td>
<td>0.466** 0.586**</td>
</tr>
<tr>
<td>6–11</td>
<td>0.412**</td>
</tr>
<tr>
<td>12–18</td>
<td>0.339**</td>
</tr>
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<td>WDS</td>
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<tr>
<td>2–5</td>
<td>0.609**</td>
</tr>
<tr>
<td>6–11</td>
<td>0.399**</td>
</tr>
<tr>
<td>12–18</td>
<td>0.391** 0.388** 0.658**</td>
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<tr>
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<tr>
<td>2–5</td>
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<tr>
<td>6–11</td>
<td>0.755** 0.543**</td>
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<tr>
<td>12–18</td>
<td>0.371** 0.366** 0.391**</td>
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<td>AnF</td>
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<tr>
<td>2–5</td>
<td>0.698** 0.622** 0.658**</td>
</tr>
<tr>
<td>6–11</td>
<td>0.309** 0.587** 0.564**</td>
</tr>
<tr>
<td>12–18</td>
<td>0.322** 0.426**</td>
</tr>
</tbody>
</table>

BT, bedtime symptoms/complaints; DS, symptoms/complaints during sleep; WDS, symptoms/complaints related to waking up from sleep; WT, symptoms/complaints at wake time; DT, symptoms/complaints during daytime; AnF, anatomical predisposing factors.

Spearman correlation test significant at p < 0.01.

Twenty-two of these children were also either undergoing or awaiting orthodontic treatment as a phase 2 post-adenotonsillectomy treatment phase.

The changes in clinical symptoms for the 99 patients with pre- and post-treatment PSG studies are outlined in Table 9. As shown, all subjects reported improvement of their SDB symptoms at clinical interviews and standardized questionnaires. Also, 43 of these 99 patients were taking medication for their clinical complaints at entry, as outlined in Table 4, while after SDB treatment only five were still taking medications.

4. Discussion

The definition of a hypopnea is very much debated. It is accepted that an AASM AHI >1 is considered abnormal in pediatric patients and studies have shown that normal children have an AASM AHI of 0. In one of our studies with normal children used to established norms, we had 60 children with AASM AHI of 0 ± 0 [3]. The result is indicative of a floor effect. In our view, the AASM criteria identifies all of the obvious, more severe cases of pediatric OSA, but will not identify a number of true OSA cases not meeting the criteria, leading to a high number of false negative test results and a low test sensitivity. This is not acceptable for a test that is potentially demanding on the families in its scheduling and preparation, is considered costly, and is used to make important health care decisions.

Table 9
Clinical symptoms reported before and after SDB treatment in the follow-up group (n = 99).

<table>
<thead>
<tr>
<th>Symptoms</th>
<th>At entry</th>
<th>At follow-up</th>
<th>Percentage of change</th>
</tr>
</thead>
<tbody>
<tr>
<td>Loud snoring</td>
<td>40</td>
<td>40.4</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Witnessed apnea</td>
<td>13</td>
<td>13</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Gasping/choking</td>
<td>13</td>
<td>13</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Excessive drooling during sleep</td>
<td>29</td>
<td>29.3</td>
<td>4 4 86.2</td>
</tr>
<tr>
<td>Sweating during sleep</td>
<td>35</td>
<td>35</td>
<td>2 2 94.3</td>
</tr>
<tr>
<td>Excessive movements during sleep</td>
<td>40</td>
<td>40.4</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Parasomnia (sleep terror, sleepwalking, sleepwalking, enuresis)</td>
<td>50</td>
<td>50.5</td>
<td>6 6 92</td>
</tr>
<tr>
<td>Bruxism</td>
<td>18</td>
<td>18</td>
<td>8 8 65.6</td>
</tr>
<tr>
<td>Disrupted nocturnal sleep</td>
<td>50</td>
<td>50.5</td>
<td>2 2 96</td>
</tr>
<tr>
<td>Difficulty falling back asleep</td>
<td>19</td>
<td>19</td>
<td>1 1 94.5</td>
</tr>
<tr>
<td>Use of hypnotics</td>
<td>11</td>
<td>11</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Hard to wake up</td>
<td>14</td>
<td>14</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Unrefreshing sleep</td>
<td>89</td>
<td>89.9</td>
<td>1 1 99.8</td>
</tr>
<tr>
<td>Morning headache</td>
<td>9</td>
<td>9</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Daytime fatigue or sleepiness</td>
<td>42</td>
<td>42</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Daytime napping</td>
<td>19</td>
<td>19</td>
<td>0 0 100</td>
</tr>
<tr>
<td>Hyperactivity-inattention</td>
<td>33</td>
<td>33</td>
<td>3 3 90.9</td>
</tr>
<tr>
<td>Use of stimulant</td>
<td>24</td>
<td>24</td>
<td>2 2 91</td>
</tr>
<tr>
<td>Headache medication</td>
<td>22</td>
<td>22</td>
<td>0 0 100</td>
</tr>
</tbody>
</table>

The table presents the number of patients and their percentage of the total studied group (n = 99), reporting symptoms before and after SDB treatment. The percentage of patients recognized by the AASM scoring criteria at entry was 9.1%, and 100% by the Stanford scoring criteria. The “percentage of change” column indicates the percentage of patients who reported improvement/complete disappearance of symptoms post treatment compared to baseline.
decisions. If clinical symptoms and clinical evaluations are more discriminative than the test itself [34,35], one may question why the test is being performed. Historically, there has been doubt about the use of thermistors to recognize abnormal breathing during sleep [36]. But the use of a nasal cannula pressure transducer has greatly improved our capability to perform analysis of the nasal airflow curve, and attended PSG allows for efficient correction of artifactual recordings. The AASM scoring system allows for both nasal cannula and thermistors to be used, but the readings obtained from each technique are different. The airflow curve is much different with the nasal cannula and the different abnormal flow patterns described in the literature with the nasal cannula cannot be identified with thermistors [37–39]. With regard to selection of our gold standard criteria for pediatric hypopnea, use of Pes has allowed for recognition of abnormal breathing during sleep in children for a long time. It has been shown that hypopneas can be associated with a decrease in effort, an important finding in children with neuromuscular diseases and in children who are at risk for hypoventilation. Abnormal events can be seen in association with “phasic REM” sleep and these hypopneas must be taken into account. Even if monitoring with Pes is considered a gold standard, we are able to obtain decent information from surface sensors with appropriately trained personnel monitoring and reading the studies [15–17]. It is true that there are some limitations with use of the nasal cannula pressure transducer such as the great difficulty in distinguishing between a hypopnea with increased effort or decreased effort during REM sleep, but the nasal cannula is clearly more sensitive than the nasal thermistor when looking at the nasal flow curve.

“Flow limitation” was defined in 1997 and is related to a flattening of the contour of the nasal flow curve obtained with a nasal cannula pressure transducer monitoring system [37]. Simultaneous use of nasal cannula and Pes showed that flow limitation was most commonly associated with an increase in inspiratory effort (as seen with the described “continuous sustained effort” pattern, Table 1). Flow limitation is often associated with snoring and may last for many minutes during nocturnal recording. Measurement of “flow limitation” is poorly represented when reduced to an “abnormal breathing event” as the duration of flow limitation may last up to 1/3 of total sleep time and cannot be appropriately represented in the scoring system. Time spent in flow limitation has been previously [16] used to describe this abnormal breathing pattern. It has been shown to be associated with an abnormal CAP rate, an associated instability of NREM sleep, and daytime symptoms [16,40]. In our study, we acknowledge that we did not calculate the amount of time spent in flow limitation, and if we had, our study may have shown a more significant breathing problem than that calculated. But the AASM scoring system does not include any guidance or option to calculate or otherwise include flow limitation. The AASM manual mentions that abnormal breathing during sleep may be detected by the presence of a “respiratory-event-related- arousal” (RERA; Table 2). However, RERAs are not included in the AASM AHI, and scoring of events is poorly defined [11].

Interesting positive and negative correlations were present in our study. Our results showed a positive correlation between AASM AHI and Stanford AHI, and all children with positive AASM AHI were positive with the Stanford scoring system. However, we also diagnosed SDB in children not identified by the AASM scoring criteria. One important source of difference between criteria is that a minimum 3% or 4% oxygen drop is required to define a non-arousal hypopnea in the AASM criteria: this is requesting an important drop in oxygen tension in a child with normal cardiopulmonary status, with high baseline SaO2 values that are located on the superior and nearly flat part of the oxyhemoglobin saturation curve, considering also presence of normal chest bellows in the supine position in normal weight individuals. This desaturation requirement selects subjects having fast drops in oxygen saturation, an uncommon finding in non-obese children having only problems in their upper airway during sleep. Investigations in children with abnormal breathing have shown sleep disturbances with shorter and different EEG patterns of arousals [40–41]. In addition, others have also argued that up to 50% of abnormal respiratory events in children are not accompanied by arousal [42].

The negative correlation between nadir of oxygen saturation and AHI has been previously noted in comparison of upper-airway-resistance-syndrome and mild apneic adult patients, particularly those with predominance or presence of only REM sleep related apneas or hypopneas [43]. Timing of onset of the abnormal breathing event in the respiratory cycle and importance of blunting of the local upper airway sensors have been suggested as potential explanations.

Short abnormal breathing events even without important oxygen saturation drops have an impact on sleep and may lead to disturbances of sleep and clinical sleep syndromes as shown by studies using the CAP scoring system [40,41] or the studies performed by the Michigan group using a computerized algorithm [44], without leading to 3% or 4% of oxygen saturation drop or transcutaneous or end-tidal CO2 increase.

The AASM scoring system recognized 19% of all the children that presented with symptoms, clinical presentation, and anatomical risk factors for OSA. As a consequence many children were left untreated, and symptomatic treatments were initiated as seen in Table 4. When looking at post-treatment results (n = 99), only 9.1% of the children had been identified by the AASM scoring system. However, 100% of the children had objective improvement with OSA treatment. When considering the lowest oxygen saturation measurement during sleep on the total treated group, there was a significant positive change of this lowest value (p = 0.001) independently of inclusion of the nine children recognized by AASM rule (n = 99) or exclusion of these nine children. Clinical complaints and symptoms reported at standardized clinical interviews and rated in validated questionnaires were improved in all 99 children post-treatment. Forty-three out of 99 children who were ingesting drugs before SDB treatment (with many children, as mentioned above, receiving several and conflicting medications such as hypnotics and stimulants) had these drugs completely withdrawn by their own physicians; this was done in 38/43 of them post SDB treatment as shown in Table 9.

The comparison shows that the AASM system rejected 81% of children with clinical symptoms, clinical evaluations, and anatomical risk factors supporting the diagnosis of SDB; these children were considered to have positive PSG with the Stanford system.

We acknowledge that we have follow-up for only 99 children, a group that consists of nine children with AASM positive PSG and 99 with Stanford positive PSG. However, SDB treatment always improved the general health of the 99 studied children, including significant decreases in reported symptoms associated with SDB and medication intake (see Table 9).

The AASM scoring guideline underscore hypopneic events in pediatric sleep disordered breathing due to the limitation set requiring a desaturation criteria and an EEG arousal >3 s. Consequently, AHI as determined by AASM criteria may deny treatment to the many patients with normal BMI. We do not affirm that current Stanford criteria are the best that can be written, but we feel that the current AASM criteria are too restrictive and limit not only appropriate recognition and treatment of children but also bias research reports.

**Conflict of interest**

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: doi:10.1016/j.sleep.2011.04.004.
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References


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