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Review Article

Accuracy of the sleep-related breathing disorder scale to diagnose obstructive sleep apnea in children: a meta-analysis

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ABSTRACT

Objectives: The main objective of this meta-analysis was to assess the accuracy of the Sleep-Related Breathing Disorder (SRBD) Scale in the diagnosis of obstructive sleep apnea syndrome (OSAS) in children.
Patients/methods: A literature search of studies comparing SRBD to polysomnography for the diagnosis of OSAS in children was performed. Risks of biases were quantified using the Quality Assessment of Diagnostic Accuracy Studies (QUADAS) tool. Analyses determined the summary receiver operator characteristic area under the curve (SROC), the pooled sensitivity (Se), the specificity (Sp), and the positive and negative likelihood ratios (LR+ and LR−). Results were graded and are expressed as means [95% confidence interval]. Post-test probabilities were computed for various populations.
Results: Eleven studies were included; and two were considered to have high risk of bias. The SROC was 0.73 [CI: 0.63; 0.82]. The combined Se, Sp, LR+ and LR− were: 0.72 [CI: 0.68; 0.77], 0.59 [CI: 0.56; 0.63], 1.74 [CI: 1.32; 2.30], 0.53 [CI: 0.39; 0.71], respectively. Sub-group analyses displayed similar results in comparison to overall results. GRADE evidence for the overall analysis was low to moderate. Finally, pre-test to post-test probabilities were estimated to be: 3.5%–1%, 50%–30% and 75%–30%, for the general population, the obese patients and the patients assigned for surgical treatment of OSAS, respectively.
Conclusions: The current meta-analysis indicates that the SRBD scale has acceptable accuracy in detecting patients with OSAS. It may be useful when evaluating patients with suspected OSAS before surgery.
Study registration: PROSPERO database (CRD42018088216).

1. Introduction

According to recent estimations, the incidence of obstructive sleep apnea syndrome (OSAS) in children ranges from 1% to 3% and increases to 60% in high-risk patients such as overweight and obese children [1–3]. Consequently, the diagnosis and management of OSAS is a daily challenge for physicians caring for children in hospitals and the community [4]. Sleep Breathing disorders in children include many conditions ranging from primary snoring to OSAS [3,5–7]. In addition, obese patients, a high-risk population for developing OSAS, might suffer from the obstructive hypoventilation syndrome, that requires specific management [8,9].

Abbreviations: OSAS, obstructive sleep apnea syndrome; SRBD, Sleep-Related Breathing Disorder; SROC, summary receiver operator characteristic area under the curve; Se, sensitivity; Sp, specificity; LR+, positive likelihood ratios; LR−, negative likelihood ratios; AHI, Apnea-Hypopnea Index; DOR, diagnostic odds ratios.

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Many physicians worldwide rely on clinical assessment – physical examination and simplified questionnaires – when diagnosing OSAS in children. This is the case even though clinical assessment has been demonstrated to be significantly inferior to the gold standard, polysomnography, whether in diagnosing OSAS or in assessing severity [1]. Reasons advocated for this practice are reduced or absent availability of polysomnography and/or inconsistency of recommendations regarding the diagnosis of OSAS [1,10,11]. Surveys among US surgeons found polysomnography to be performed in 10% of children before tonsillectomy [12,13]. Inaccurate diagnosis leads to mismatched therapy and potentially serious consequences in OSAS patients undergoing surgery [14].

At the beginning of this century, Chervin and collaborators [15] developed the Sleep-Related Breathing Disorder (SRBD) Scale, deriving their scale from the pediatric sleep questionnaire (a 22-item SRBD questionnaire with a positive classification defined as the presence of more than 33% of positive response). Using an Apnea-Hypopnea Index (AHI) > 5 to define the presence of OSAS during gold standard polysomnography comparison, they found this questionnaire to exhibit a sensitivity and specificity of 85% and 87%, respectively, in the construct cohort; and 81% and 87%, respectively, in the validation cohort. A meta-analysis of clinical studies in this era has found moderate sensitivity and specificity of mixed clinical signs in the diagnosis of OSAS in children in comparison to polysomnography [16]. As an example, Van Someren et al., found the doctor's examination, to exhibit a sensitivity and specificity of 59% and 73%, respectively when compared to polysomnography [17] and A similar result (sensitivity 55%) was also found in another review of the reliability of history and physical examination in the diagnosis of OSAS [18].

The main objective of this meta-analysis was to determine the accuracy of the SRBD Scale compared to polysomnography, in the diagnosis of the OSAS in children.

2. Material and methods

2.1. Bibliographic search and analysis

We conducted this meta-analysis according to the Cochrane Handbook for Systematic Reviews and the PRISMA guidelines [19]. This study was registered in the PROSPERO database (CRD42018088216). No deviation from the registered protocol was performed.

Literature databases included Pubmed, the Embase Cochrane central register of controlled trials, and the clinical trials register. The following keywords “children or infant” and “polysomnography” were associated with “paediatric sleep questionnaire” or “pediatric sleep questionnaire” or “Sleep-Related Breathing Disorder Scale.” Language search was not restricted. The pediatric population was defined as <18 years old. The date of the last assessment was July 2018. All searches were performed by an individual participant in the study.

Articles obtained from queries were independently analyzed by eight senior anesthesiologists-the list is available in the author's section, and the results were checked twice. Disagreements between readers were resolved by consensus.

Extracted data consisted of: age of patients, populations in which the study was performed (specific medical conditions or populations), the risk of bias as defined by QUADAS tool analysis (see below), sample size, polysomnographic criteria defining the diagnosis of OSAS in each article, and table 2 × 2 items: true positives, false positives, true negatives and false negatives. Consequently, articles with the following criteria were included in the analysis: intervention: [1] articles that assessed the accuracy of the SRBD Scale derived from the pediatric sleep questionnaire as developed and validated by Chervin RD and collaborators [15], [2] articles that compared SRBD with polysomnography in the diagnosis of obstructive sleep apnea syndrome, and [3] articles that provided sufficient information to construct a 2 × 2 contingency table employing a Sleep-Related Breathing Disorder Scale result of 0.33 or 33% of positive responses as a definition of OSAS. Meeting abstracts were not included in this meta-analysis, to allow a full determination of the quality of included studies. The presence of potential bias was assessed using the Quality Assessment of Diagnostic Accuracy Studies (QUADAS) tool [20], a specific validated tool for the quality assessment of diagnostic accuracy studies. Areas of the tool are: (1) Patient selection: described methods of patient selection, described included patients (prior testing, presentation, intended use of index test and setting); (2) Index test: described the index test and how it was conducted and interpreted; (3) Reference standard: described the reference standard and how it was conducted and interpreted; (4) Flow and timing: described any patients who did not receive the index test(s) and/or reference standard or who were excluded from the 2 × 2 table (refer to flow diagram), described the time interval and any interventions between index test(s) and reference standard.

The primary outcome of the study was the accuracy of the SRBD scale (sensitivity and specificity) in diagnosing OSAS in children. A good accuracy was considered if the sensitivity was ≥95% [21]. Secondary outcomes were: the summary receiver operator characteristic area under the curve, the positive/negative likelihood ratios, diagnostic odds ratios (DOR) and the pre-test to post-test probability in the general population of certain medical conditions, obese patients and those referred for surgical management of OSAS.

2.2. Statistical analysis

The meta-analysis was performed using a bivariate meta-analysis model [22,23] to calculate the following performance parameters: pooled sensitivity, specificity, positive/negative likelihood ratios, and diagnostic odds ratios (DOR). For each parameter, the I^2 test was used to assess the heterogeneity of results [24]. According to the Cochrane handbook an $I^2 > 40\%$ indicates the presence of heterogeneity (moderate: 40%–60%; considerable: 75%–100%).

The summary receiver operator characteristic (SROC: that plotted sensitivity versus 1-specificity) curve of the overall result and subgroup analyses was constructed, and the mean and 95% confidence intervals of the area under the curve were determined [25]. Additional graphical representations were added to the SROC concerning the overall result: the 95% confidence region and the 95% prediction regions. The confidence region evaluates the precision of estimation, and the prediction region evaluates variations between included studies. Publication bias was assessed with Deek's funnel plot: a linear regression test between the natural logarithm of the OR against the root of effective sample size (defined as $4 \times (N_{test} \times N_{control})/N_{total}$) [26,27].

Subgroup analyses were planned for the following factors: quality of included studies (analyses including studies with no high-risk of bias detected), health conditions or particular situations (examples: surgical population, obesity) and polysomnographic criteria defining the presence of OSAS (if different between studies). Subgroups were considered if recorded in at least two studies that were included in the analysis. When two or more samples were analyzed within one study, each sample was considered as an individual study for the purpose of analysis.

In order to assess the usefulness of the SRBD in daily practice, we determined the post-test probability of OSAS in three conditions using Fagan's nomogram: in the general population (pre-test

probability: 2%) [1], in patients at risk of OSAS referred to a specialized centers for further investigation (clinical diagnosis with a pre-test probability: 55%) [18] and in obesity (pre-test probability: 36%) [28–30].

Finally, the summary of overall analyses and secondary analyses including only studies without high-risk of bias was included in the GRADE analysis. To assess the overall quality of evidence for each outcome (pooled accuracy parameters), we downgraded the GRADE ranking by one level from 'high quality' for each of the following observations: high risk of bias, indirectness of evidence, serious inconsistency (heterogeneity as defined above) and potential publication bias [31–33]. The large confidence interval was not considered as a reason for downgrading evidence given that no reference could be established for the large confidence interval. However, the value of sensitivity <95% and/or a positive likelihood ratio > 5–10 and/or a negative likelihood ratio <0.1 were considered as a serious inconsistency mandating a one-level downgrading [21].

Statistical analyses used the following packages: Meta-Disc (version 1.4, Unitad de Bioestadística Clinica, Hospital Romon y Cajal. Madrid. Spain) [34] for computing accuracy parameters and SROC curves, the R package (R version 3.2.3 The R Foundation for Statistical Computing; Library Mada) for computing the 95% confidence and prediction regions, the RevMan 5 meta-analysis tool (Version 5, RevMan 5.3, The Cochrane Collaboration, Oxford, United Kingdom) for editing the QUADAS tool, the GraphPad software (version 5, GraphPad Software, San Diego, California, USA) for the Deek's funnel plot and the GRADE PRO online program for GRADE Analysis.

Data were expressed as a mean [95% confidence interval]; the level of significance was defined as 0.05.

3. Results

Search criteria identified 103 potential appropriate publications and 25 potential studies. Once inclusion and exclusion criteria were applied, 11 studies including 1162 patients were selected for meta-analysis (flow-chart Fig. 1) [15,35–44]. Table 1 displays the description of studies included. Two studies were found with two independent samples each (Table 1) [15,44]. Those two studies used the same polysomnographic thresholds to define the presence of OSAS (Table 1). Accordingly, analyses were carried with results from 13 patient samples. No study was discarded or translated because of non-English language, and all studies were published in peer-reviewed journals.

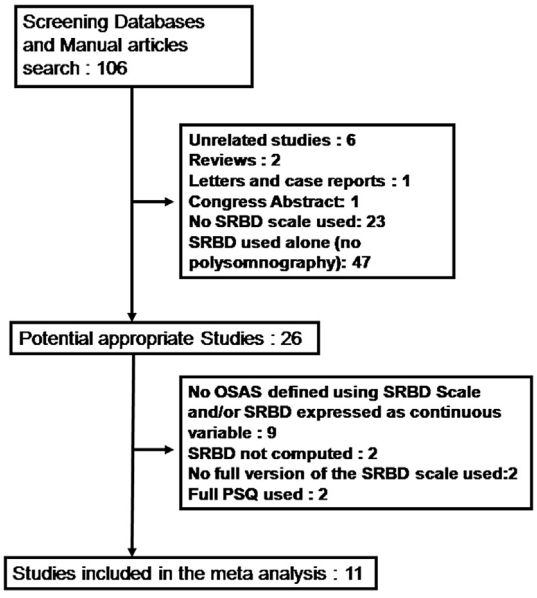

Fig. 1. Flowchart of included studies.

Study quality averaged "good" with either no risk or undetermined risk of bias (Fig. 2). Two studies were assessed to be high risk for bias: one because of patient selection based on non-random criteria [44], and the other because of the lack of a clear definition of the diagnostic AHI threshold for OSAS during polysomnography [36]. The recruitment of patients in the original publication of Chervin et al., [15] was assessed by our authors as an undetermined risk. The investigated population mixed patients referred for a suspicion of OSAS and patients not suspected to carry this condition. However, each subgroup appears to have been randomly selected. All included studies evaluated the SRBD scale concomitant with performing polysomnography.

3.1. Overall results

The combined sensitivity (Supplemental file 1) and specificity (Supplemental file 2) were (Table 2): 0.73 [0.68; 0.77] (I^2 = 69.4%), 0.59 [0.56; 0.63] (I^2 = 89.8%). The Positive likelihood ratio and negative likelihood ratio were (Table 2) 1.74 [1.32; 2.30] (I^2 = 83.4%), 0.53 [0.39; 0.71] (I^2 = 60%), respectively. The Diagnostic Odds Ratio (Table 2) was found to be 3.64 [2.02; 6.56] (I^2 = 73.2%). The area under the curve of the SROC analysis was 0.73 [0.63; 0.82] (Fig. 3, Table 2). Graphically, the 95% confidence region was limited and above the identity line, whilst the 95% prediction region was much larger and crossed the identity line (Fig. 3).

3.2. Study bias analyses (Table 2)

Excluding studies with high-risk of bias [36,44] (see above and Fig. 2 for details) the study bias analysis produced results similar to the overall analysis but with higher specificity and a higher positive likelihood ratio (Table 2).

3.3. Subgroup analyses (Table 2)

Using the methodology described above, we defined subgroups as obese or overweight patients, the definition of OSAS as AHI > 5/hour and definition of OSAS as AHI ≥ 1/hour or ≥1.5/hour. Results in obese patients (three studies with three patient samples) and studies defining OSAS as AHI ≥ 1/hour or ≥1.5/hour (five studies including six patient samples), were similar when compared to overall results. Focusing on studies defining OSAS as an AHI > 5/hour (four GRA studies with five patient samples), resulted in improved accuracy parameters, and with the exception of specificity that remained unchanged when compared with overall results.

3.4. Grade analysis

GRADE evidence for the overall analysis was downgraded three times as two studies with potential high risk of bias were included; heterogeneity was found in results (I^2 = 69.4% and 89.8% for sensitivity and specificity, respectively, Table 2) and sensitivity was <95% (in addition to a positive likelihood ratio >5–10 and a negative likelihood ratio <0.1). Overall the quality of evidence was assessed as low (Table 3). Excluding studies with high-risk of bias [36,44] increased the GRADE evidence rating to moderate (Table 3).

Table 1
Description of included studies. Studies with more than one subgroup are displayed as individual studies (Studies of Chervin 2000 [15] and Ward 2016 [44] were duplicated because each included two cohorts that were entered in the analyses as separate studies). RDI: respiratory disturbance index; AHI: Apnea-Hypopnea Index.

Author, Date	Age	Population	N	Polysomnography criteria	Sensitivity	Specificity
Alonso-Alvares, 2014 [35]	3 to 14	Obesity	248	RDI ≥ 3/h of total sleep time	0.545	0.691
Amin, 2015 [36]	0 to 18	Kidney Failure	19	Undetermined	0.28	0.58
Bertran, 2014 [37]	0 to 15	Various	83	AHI ≥ 1	0.71	0.52
Carno, 2008 [38]	8 to 17	Overweight and obese	151	AHI > 5	0.84	0.23
Chan, 2012 [39]	2 to 18	Various	102	AHI > 1.5	0.5	0.55
Chervin, 2000 [15]	2 to 18	Various	81	AHI > 5	0.81	0.87
Charvin, 2000 [15]	2 to 18	Various	81	AHI > 5	0.85	0.87
Chervin, 2007 [40]	5 to 13	AT preoperative	105	AI ≥ 1	0.78	0.72
Cielo, 2014 [41]	2 to 18	Craniofacial disorders	83	AHI > 5	0.57	0.48
Ishman S, 2016 [42]	0 to 18	Obesity	45	AHI > 5	0.86	0.38
Uyan, 2016 [43]	3 to 18	Chronic Lung Disease	21	AHI > 1	0.5	0.82
Ward, 2017 [44]	6 to 11	Juvenile Idiopathic Arthritis	68	AHI > 1,5	0.86	0.3
Ward, 2017 [44]	6 to 11	Control patient	75	AHI ≥ 1,5	0.62	0.42

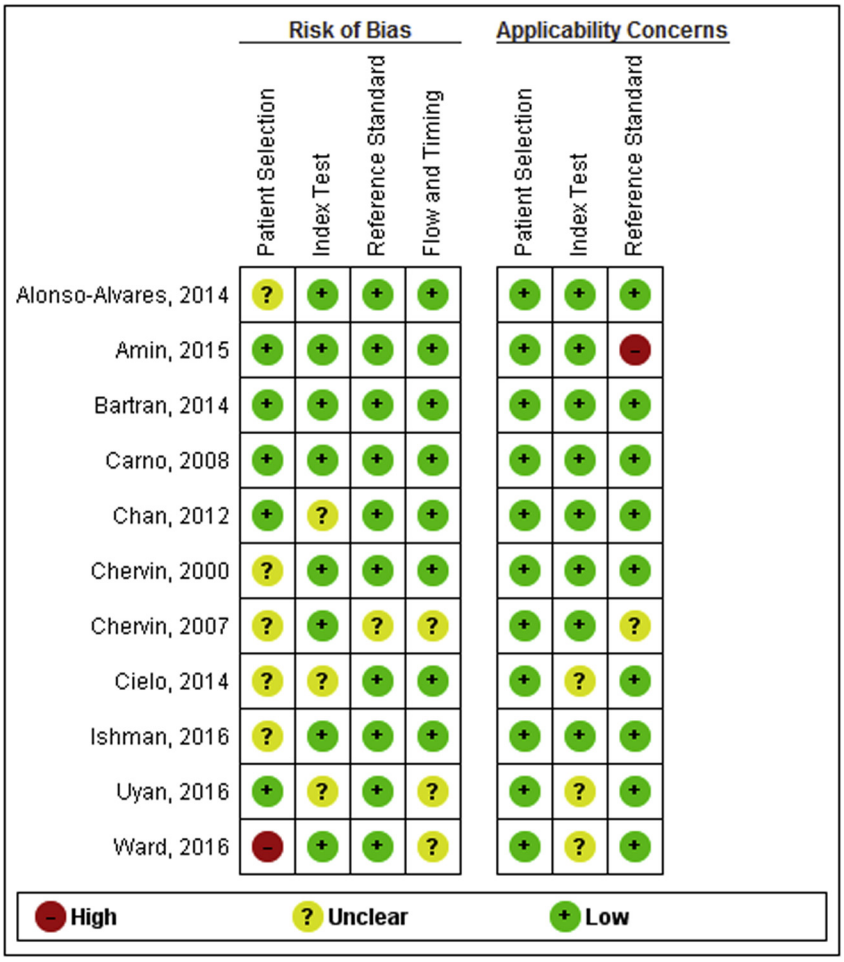

Fig. 2. Bias analyses of included studies using the QUADAS tool. Areas of the tool are: (1) Patient selection: Described methods of patient selection, description included patients (prior testing, presentation, intended use of index test and setting), (2) Index test: described the index test and how it was conducted and interpreted, (3) Reference standard: described the reference standard and how it was conducted and interpreted and (4) Flow and timing: described any patients who did not receive the index test(s) and/or reference standard or who were excluded from the 2 × 2 table (refer to flow diagram), described the time interval and any interventions between index test(s) and reference standard.

3.5. Publication bias analysis

According to the method described above, no correlation was found between the natural logarithm of the OR against the root of effective sample size (Fig. 4). Accordingly, publication bias was excluded.

3.6. Usefulness of the SRBD scale in three common situations

Using Fagan's nomogram, we determined the post-test probability in three common situations with an estimated pre-test probability (Fig. 5). Results using overall analyses indicate that in the general population-Fig. 5A, SRDB Scale results of >0.33 and

Table 2
Subgroup analyses. Studies with more than one subgroup are displayed as individual studies. AHI: Apnea-Hypopnea Index.

	Overall (Number of studies = 13; Number of individuals = 1162)	No high risk of bias (Number of studies = 10; Number of individuals = 1000)	Obese (Number of studies = three; Number of individuals = 444)	AHI ≥ 1 (Number of studies = six; Number of individuals = 578)	AHI > 5 (Number of studies = five; Number of individuals = 441)
Area under the curve	0.73 [0.63; 0.82]	0.74 [0.63; 0.85]	0.65 [0.58; 0.72]	0.67 [0.54; 0.80]	0.84 [0.69; 0.99]
Sensitivity	0.73 [0.68; 0.77] I^2 = 69.4%	0.71 [0.68; 0.76] I^2 = 74.4%	0.72 [0.65; 0.78] I^2 = 90.5%	0.71 [0.64; 0.79] I^2 = 57.4%	0.81 [0.74; 0.86] I^2 = 52.9%
Specificity	0.59 [0.56; 0.63] I^2 = 89.8%	0.62 [0.58; 0.66] I^2 = 90.4%	0.56 [0.49; 0.62] I^2 = 95.3%	0.54 [0.48; 0.60] I^2 = 81%	0.58 [0.52; 0.64] I^2 = 95%
Positive likelihood ratio (LR+)	1.74 [1.32; 2.30] I^2 = 83.4%	1.91 [1.34; 2.73] I^2 = 87.1%	1.36 [0.96; 1.93] I^2 = 79.2%	1.49 [1.10; 2.04] I^2 = 65%	2.18 [1.11; 4.29] I^2 = 93.1%
Negative likelihood ratio (LR-)	0.53 [0.39; 0.71] I^2 = 60%	0.51 [0.36; 0.72] I^2 = 69.1%	0.65 [0.51; 0.83] I^2 = 0%	0.58 [0.40; 0.87] I^2 = 50.1%	0.40 [0.19; 0.83] I^2 = 77.5%
Diagnistic odd ratio (DOR)	3.64 [2.02; 6.56] I^2 = 73.2%	4.12 [2.06; 8.25] I^2 = 79.1%	2.33 [1.50; 3.62] I^2 = 0%	2.74 [1.35; 5.54] I^2 = 54.8%	5.82 [1.39; 24.38] I^2 = 87.6%

<0.33 indicated post-test probabilities of the diagnosis of OSAS of 3.5% and 1%, respectively. In obese patient (Fig. 5B) subgroups, SRBD Scale results of >0.33 and <0.33 indicated post-test probabilities of the diagnosis of OSAS of 50% and 30%, respectively. Finally, in patients referred for surgery with a high suspicion of OSAS (sub-group AHI > 5/h), SRBD Scale results of >0.33 and <0.33 indicated post-test probabilities of 75% and 30%, respectively (Fig. 5C). Values of positive and negative likelihood ratios used for computing post-test probability in this population were those obtained from the subgroup of patients with an AHI >5 (defining moderate to severe OSAS) [45].

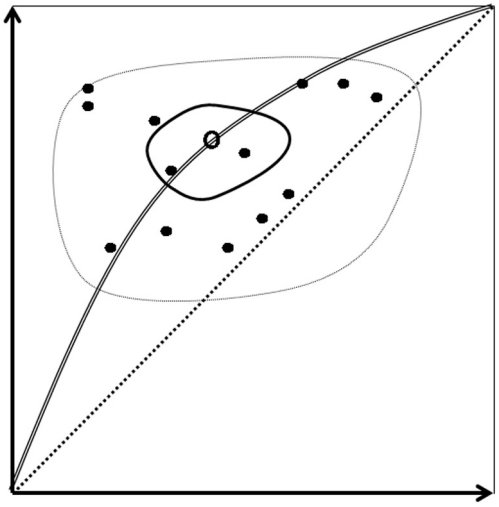

Fig. 3. The summary receiver operator characteristic curve (displayed as a double line), its 95% confidence region (full line area) and its 95% prediction regions (dashed line area). The confidence region evaluates the precision of estimation, and the prediction region evaluates variations between included studies.

4. Discussion

The main finding of this meta-analysis can be summarized as follows: according to accuracy parameters, SROC analysis and the

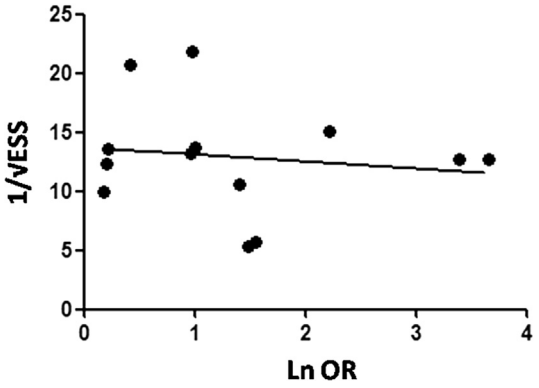

Fig. 4. Deek's funnel plot for the detection of bias in meta-analysis. X-axis natural logarithm of Odd Ratio of individual studies, Y-axis the effective sample size of each study. Ln OR: natural logarithm of the OR, √ESS: the root of effective sample size (defined as 4 × (Ntest × Ncontrol)/Ntotal).

Table 3
Grade analyses of the levels of clinical evidence.

No of studies (No of patients)	Study design	Factors that may decrease certainty of evidence					Test accuracy
		Risk of bias	Indirectness	Inconsistency	Imprecision	Publication bias	
11 studies (1162 patients)	cross-sectional (cohort type accuracy study)	serious[a]	not serious	serious[b,c]	not serious	strong association	⊕⊕◯◯ Low
Nine studies (936 patients)	cross-sectional (cohort type accuracy study)	not serious	not serious	serious[b,c]	not serious	strong association	⊕⊕⊕◯ Moderate

[a] Two studies with high-risk of bias.
[b] Heterogeneity of results.
[c] Sensitivity <95% or a positive likelihood ratio > 5–10 or a negative likelihood ratio <0.1.

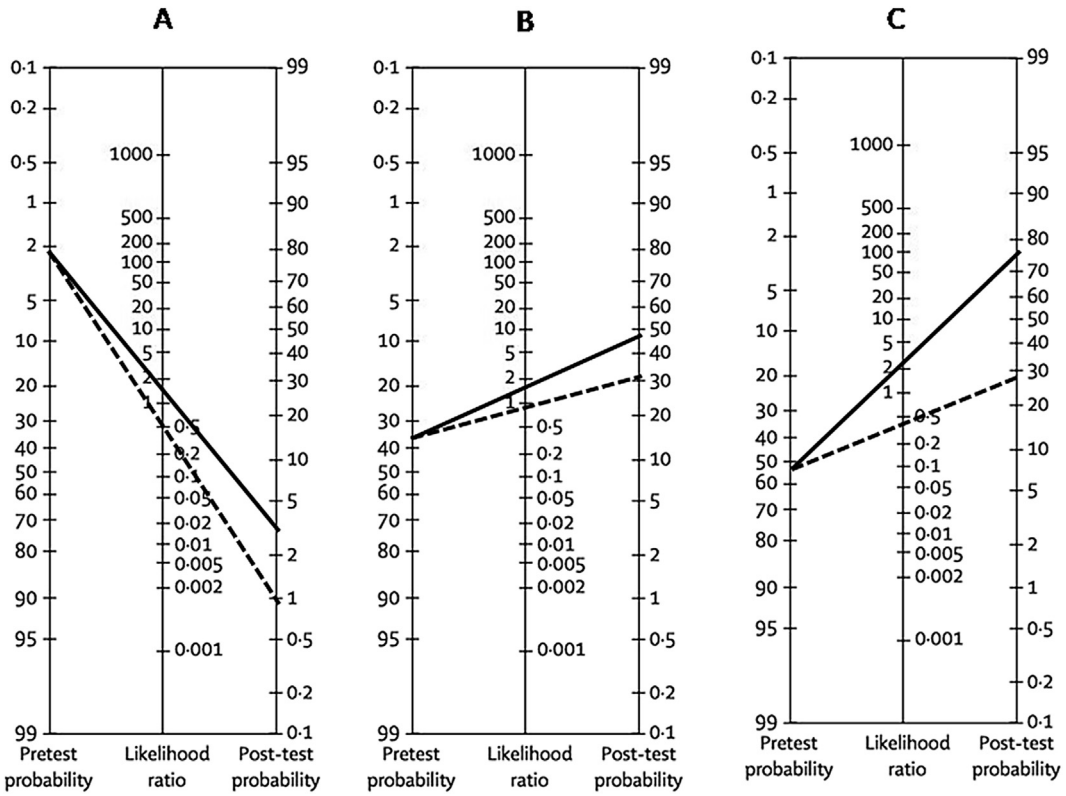

Fig. 5. Fagan's nomogram for three common situations: A: general pediatric population, B: obese patients and C: patients with a clinical diagnosis of obstructive apnea syndrome. Full lines: post-test positive probability, dashed lines: post-test negative probability.

extent of the predictive area, the SRBD Scale derived from the pediatric-sleep questionnaire was found to be moderately accurate in the diagnosis of OSAS in children. GRADE assessment of the quality of evidence resulted in low to moderate quality results, and no publication bias was detected.

According to the results displayed by overall analyses, the sensitivity of the SRBD scale was slightly lower in comparison to the original validating study performed by Chervin et al., [15] (0.73 [0.68; 0.77] versus 0.81 [0.62; 0.94], respectively). Specificity diverged more markedly, however (0.59 [0.56; 0.63] versus 0.87 [0.75; 0.95], respectively). This indicates a reduced ability of the SRBD scale to detect patients without OSAS when the questionnaire is negative. This result is likely to be explained by the heterogeneous population included in this meta-analysis and is supported by the heterogeneity of results observed in both overall and subgroups analyses and by the extensive surface of the prediction region that evaluates variations between included studies. Another explanation is the mismatch between the polysomnographic parameters used to define the presence of OSAS in the original publication of Chervin et al., (AHI > 5/hour) and those of the current meta-analysis, which combined studies using a different polysomnographic definition of OSAS (both AHI > 5/hour and AHI ≥ 1/hour). However, variations in polysomnographic diagnostic thresholds are unlikely to have significantly affected results given that the SRDB scale specificity results were similar for the overall analysis and for the subgroup of studies defining OSAS with an AHI > 5/hour (Table 2). Finally, the recruitment of patients in the Chervin's study [15], they mixed patients with high risk and low risk of OSAS, may explain discrepancies in results; most studies included in this meta-analysis randomly selected patients from predetermined populations. This is highly supported by the fact that the prevalence of OSAS in the Chervin's cohorts was 50% while it was lower (41%) and more heterogeneous (10%–93%) when all included studies were considered.

The SRBD scale exhibits a moderate accuracy given the SROC analysis and accuracy parameter results (sensitivity < 95%, the positive likelihood ratio > 5–10 and the negative likelihood ratio < 0.1). Furthermore, it was comparable in predicting OSAS in pooled results, in patients with moderate to severe OSAS (AHI > 5/hour) and in patients at high risk of OSAS (such as obese or overweight ones) (Table 2). Moreover, the number of studies (and patients) included for computing subgroups were limited (Table 2). Finally, GRADE quality of evidence was assessed as low to moderate. Considering all those results, one may question the routine use of the SRBD scale; the use of the pooled sensitivity and specificity are of little interest when focusing on particular subgroups. Accordingly, the determination of post-test probabilities in different population was important to assess the potential usefulness of this scale.

Using the results of this meta-analysis, we determined the post-test probabilities of OSAS in children in some “usual” clinical situations. A negative questionnaire in the general pediatric population excludes the diagnosis (5%–1.5% for pre and post-probabilities, respectively). However, given that numerous OSAS risk factors that increase pre-test probability are present in the general population (male gender, ethnicity…), the use of the SRBD scale appears more appropriate in epidemiological studies of the general population, rather than individual patient screening. The SRBD scale was found to be irrelevant for individual patient screening in all obese patients (Fig. 5) given that post-test probabilities were inconclusive (70% and 40% when results of the SRBD scale were > or <0.33). This is unsurprising given the high pre-test probability for OSAS in obese patients (60%). Yet, considering the typical scenario commonly encountered by surgeons deciding whether to proceed or not with

tonsillectomy when facing children with suspicion of OSAS, a positive SRBD scale result increases the probability of disease from 55% to 75%, while a negative result decreases it to 30%. Where polysomnography is not available, surgery would be expected to be more appropriate and hypothetically more beneficial in children with a 75% probability of having an OSAS than in those with a 55% probability. Moreover, fewer unnecessary tonsillectomies will be performed; this is pertinent because, although infrequent, serious complications do occur [14]. Alternatively, one can also imagine the SRBD scale being used as a screening tool; then, for a more accurate indication, proceeding to polysomnography where the latter is not easily available. Nocturnal oximetry is an interesting alternative to polysomnography proposed in order to overcome the unavailability of polysomnography. A recent meta-analysis summarizing the accuracy of this method in the diagnosis of OSAS (defined as an AHI > 1/hour) found a summary sensitivity and specificity of 77.7% and 88.9%, respectively [46]. Although values are still far from the 95% objective [21], one can hypothesize a complementary effect combining nocturnal oximetry with an SRDB questionnaire.

One of the major concerns in using the SRBD scale is the absence of any indication of OSAS severity. The questionnaire was not equipped to identify patients with perioperative respiratory risks. This is supported by the sensitivity accuracy parameters for moderate to severe OSAS (AHI > 5/hour) identified in the current study, that were slightly better than those for mild to severe OSAS (AHI $\geq$ 1/hour). Specificity accuracy parameters were comparable across moderate to severe and mild to severe patient groups (Table 1). Accordingly, this questionnaire, in contrast to polysomnography, cannot be used to assess perioperative risk in children undergoing tonsillectomy or to plan appropriate perioperative management such as ambulatory surgery and opioid administration [14]. One recent study further derives a specific questionnaire from the SRBD scale, attempting to predict a higher risk of perioperative respiratory complications [47]. However, for the time being, the lack of follow up validation studies of this questionnaire represents a serious question mark over its routine clinical use.

The current meta-analysis suffers some limitations that must be taken into account. First, results were heterogeneous, both in overall results and subgroup analyses. This suggests that despite the number of patients included and the provision to homogenate the analyses, some other conditions still differ between studies such as health condition, age, gender, ethnicity, facial and bony abnormalities, tonsil size, and neurological disorders. One other significant limitation of the study is the small number of studies available for analysis that probably impacted upon analysis quality. However, no publication bias was observed, which indicates a low risk of negative unpublished studies. The current study explored the accuracy of the SRBD scale in the diagnostic of OSAS. However, physicians should be aware of the fact that disordered sleeping includes a wide spectrum of syndromes not explored in the current study [4]. These include primary snoring defined by an AHI <1/hour which is considered an indication for surgery (tonsillectomy) when associated with systemic or behavioral complications [5,6]. Alternatively, the obstructive hypoventilation syndrome associated with obesity is a condition usually associated with OSAS that mandates non-surgical management such as nocturnal positive-pressure support and weight loss [8,9,48]. Physicians should be aware of these conditions that limit the usefulness of the SRBD scale. Finally, one of the most important limitations of the study is the relatively few clinical circumstances in which the SRBD scale appeared to be relevant, as demonstrated by the pre-test/post-test analyses. Yet, as stated in the introduction, the main problem that is facing physicians in many parts of the world is the decision whether or not to propose surgery for patients without polysomnographic results; our analysis strongly suggests that the SRBD might be of great help in this particular situation.

In conclusion, the current meta-analysis indicates that the SRBD scale has a more acceptable accuracy in detecting patients with OSAS compared to clinical examination. It is potentially useful as an epidemiological tool and to identify children appropriate to undergo complementary evaluation (polysomnography or nocturnal saturation) before tonsillectomy. Future studies combining this questionnaire with other simple diagnostic tools such as nocturnal oximetry would represent a suitable option in the diagnostic of OSAS in children.

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Authors contribution

Daphné Michelet: conceptualized and designed the study, selected articles, evaluated articles, collected data, corrected the manuscript and approved the final manuscript as submitted.

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Christopher Brasher: conceptualized and designed the study, verified statistics, drafted the initial MSS, corrected the manuscript and approved the final manuscript as submitted.

Souhayl Dahmani: conceptualized and designed the study, designed the data collection instruments, carried out the initial analyses and verified statistics, drafted the initial manuscript, corrected the manuscript and approved the final manuscript as submitted.

Conflict of interest

None declared.

The ICMJE Uniform Disclosure Form for Potential Conflicts of Interest associated with this article can be viewed by clicking on the following link: https://doi.org/10.1016/j.sleep.2018.09.027.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.sleep.2018.09.027.

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