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CLINICAL REVIEW

Diagnostic accuracy of screening questionnaires for obstructive sleep apnea in children: A systematic review and meta-analysis

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SUMMARY

This systematic review and meta-analysis evaluated the diagnostic accuracy of screening questionnaires for pediatric obstructive sleep apnea (OSA).

Studies comparing any questionnaire with polysomnography for OSA detection in subjects aged ≤18 y were considered eligible for qualitative analysis. The quality assessment of diagnostic accuracy studies (QUADAS-2) tool was used for bias assessment. Only questionnaires adopted by at least four studies using the currently accepted diagnostic threshold of apnea–hypopnea index (AHI) ≥1 were included for further selective quantitative analyses. A bivariate meta-analysis was performed to calculate sensitivity, specificity, positive and negative likelihood ratios, diagnostic odds ratio; summary receiver operator characteristic curves were constructed.

37 studies (20 questionnaires) were eligible for qualitative analysis; none were considered of low quality. Among these articles, 13 studies and two questionnaires (sleep-related breathing disorder scale of the pediatric sleep questionnaire (SRBD-PSQ) and OSA-18) satisfied the criteria for quantitative synthesis. SRBD-PSQ had higher sensitivity (0.76) than OSA-18 (0.56), while OSA-18 exhibited higher specificity (0.73) than SRBD-PSQ (0.43).

SRBD-PSQ performed well and was the most sensitive screening questionnaire using the diagnostic threshold of AHI ≥1 for pediatric OSA. However, further well-designed studies are still required to assess the role of SRBD-PSQ in real-world clinical populations.

Introduction

Obstructive sleep apnea (OSA) is among the most severe form of sleep-disordered breathing in children; it is characterized by "prolonged partial upper airway obstruction and/or intermittent complete obstruction (obstructive apnea) that disrupts normal ventilation during sleep and normal sleep patterns" [1,2]. Signs and symptoms may include snoring, labored/obstructed breathing, or daytime consequences such as sleepiness and hyperactivity [2].

Prevalence rates of OSA in children range from 1.2% to 5.7% [3]. If untreated, pediatric OSA may be associated with sequelae such as behavioral disturbances, hyperactivity, learning difficulties, growth delay and possible but long-term negative effects on cardiovascular health [4]. Therefore, OSA is a relatively common problem that can be encountered by most clinicians who treat children; early diagnosis and treatment are mandatory given the potential benefits on child's development, school performance, cognitive abilities and social interactions.

Polysomnography (PSG) is the current gold standard diagnostic test for OSA and the apnea-hypopnea index (AHI, i.e., the number of obstructive and mixed apneas and hypopneas per hour of total sleep time) is the PSG parameter most commonly reported to discriminate the presence or the severity of OSA. The diagnostic criteria for pediatric OSA require either AHI≥1, or obstructive

Abbreviations: AHI, apnea–hypopnea index; CI, confidence interval; DOR, diagnostic odds ratio; OSA, obstructive sleep apnea; PRISMA, preferred reporting items for systematic review and meta-analysis; PSG, polysomnography; PSQ, pediatric sleep questionnaire; QUADAS, quality assessment of diagnostic accuracy studies; SRBD-PSQ, sleep-related breathing disorder scale of the pediatric sleep questionnaire; SROC, summary receiver operator characteristic.

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hypoventilation manifested by peripheral arterial carbon dioxide >50 mmHg for more than 25% of sleep time, coupled with snoring, paradoxic thoracoabdominal movement, or flattening of the nasal airway pressure waveform [3].

Since PSG is expensive and not widely available, screening questionnaires may offer a simple and cost-effective alternative to PSG for the identification of children at high risk of OSA who will benefit from referral to a sleep specialist. Questionnaires may also help to triage and prioritize the long wait lists at sleep specialists for children already referred.

Several primary studies attempted to evaluate the ability of pediatric screening questionnaires to diagnose OSA using different PSG criteria to define the presence of the disease [5–21]. An AHI≥5 was most commonly chosen as the diagnostic cutoff instead of the children cutoff value of AHI≥1 [7,8,11], but not exclusively (1.5 [9,10,12,13], 2 [5,15], 3 [16]). Even different PSG parameters have been used (e.g., respiratory disturbance indices [19], obstructive index [20], obstructive apnea index [21]). However, to correctly evaluate the diagnostic accuracy it is necessary to make comparisons with the same reference standard parameter. Systematic reviews on this topic are, therefore, essential. Previous systematic reviews investigated questionnaires diagnostic capability investigating only one questionnaire [22], without conducting a meta-analysis [23], or combining studies with different index tests or different AHI thresholds [22,24,25].

The aim of this systematic review and meta-analysis was to conduct a qualitative and a further selective quantitative analysis in order to provide an up-to-date evaluation of the diagnostic accuracy of pediatric screening questionnaires and compare them with PSG as the reference standard for the diagnosis of OSA in children.

Methods

This systematic review was conducted according to Cochrane handbook for systematic reviews of diagnostic test accuracy [26] and to the preferred reporting items for systematic review and meta-Analysis (PRISMA) statement [27]. This study was registered in the PROSPERO database (registration number CRD42019134919).

Eligibility criteria

In the first step of qualitative analysis all types of studies, both prospective and retrospective, were considered eligible according to the following inclusion criteria: 1) use of any parent answered questionnaire for the screening of OSA in children, 2) participants under 18 y of age, 3) PSG employed as the reference standard to confirm the diagnosis of OSA (as recommended by the American Academy of Sleep Medicine [28]).

Exclusion criteria were: 1) use of any other diagnostic tests (e.g., clinic exams, pulse oximetry, electrocardiography, laboratory testing), 2) lack of data to measure sensitivity, specificity, diagnostic odds ratio, positive and negative likelihood ratio of screening questionnaires, and 3) exclusive evaluation of other sleep related problems (e.g., sleepiness, insomnia, narcolepsy, nighttime walking, difficulty to wake up).

The studies considered eligible for the qualitative analysis were further selected for subsequent quantitative analyses according to the following inclusion criteria: 1) use of the currently accepted diagnostic threshold of AHI ≥1, 2) questionnaires adopted by at least four studies. Exclusion criteria were: 1) low quality studies and 2) small sample size (less than 30 participants).

Search methods

The following electronic bibliographic databases were searched without time or language restrictions: PubMed, Scopus, Web of Science and The Cochrane Library. The following terms and keywords were used: obstructive sleep apnea, sleep, child, children, adolescent, questionnaire, accuracy, validity, evaluation. Reference lists of eligible studies, key journals, trial registers were also searched.

The searches were re-run just before the final analyses and further studies retrieved for inclusion (last updated search was in May 2020). The search strategy for PubMed is shown in Table S1.

Study selection

Titles and abstracts were screened independently by two review authors in order to exclude duplicates and irrelevant articles. The full texts of potentially eligible studies were retrieved and examined in accordance with the eligibility criteria by two review authors in the first step of qualitative analysis and, also, in the second step of further selective quantitative analysis. Any disagreement was resolved through discussion. Where resolution was not be possible, a third reviewer was consulted.

Qualitative analysis

Data extraction and management

A standardized, pre-piloted form was used to extract data from the included studies. Extracted information included study setting, study population, participant demographic and baseline characteristics, polysomnographic criteria for the diagnosis of OSA, prevalence, sensitivity, specificity, positive predictive value, negative predictive value, positive likelihood ratio, negative likelihood ratio, diagnostic odd ratio (DOR), true positives, false positives, true negatives and false negatives. Two review authors extracted data independently. Discrepancies were identified and resolved through discussion (with a third author where resolution was not be possible). No attempt was made to contact the study authors for missing data.

Quality assessment

Two review authors independently assessed the quality of studies included in the qualitative synthesis using the quality assessment of diagnostic accuracy studies (QUADAS-2) instrument [29].

This tool comprises four key domains that discuss the risk of bias associated with patient selection (methods of patient selection, description of included patients), index test (how it was conducted and interpreted), reference standard (how it was conducted and interpreted), flow and timing (the time interval and any interventions between index test and reference standard). The first three domains are also assessed in terms of concerns about applicability. Assessment of risk of bias and concerns about applicability are treated as distinct components of quality assessment and judged as “Low”, “High”, or “Unclear”.

Any disagreement was resolved by discussion, with involvement of a third review author where resolution was not possible.

Quantitative analysis

Data synthesis

Bivariate meta-analysis was performed to calculate the following performance parameters: sensitivity, specificity, positive/negative likelihood ratios, and DOR. If data were unavailable in the included studies, we computed them using the available ones. The summary receiver operator characteristic (SROC) curves were constructed, and the 95% confidence region and the 95% prediction regions were added.

For the questionnaires included in the quantitative analysis, the studies using a diagnostic threshold of AHI≥5 (i.e., the currently accepted diagnostic threshold for moderate-severe OSA in children) were also evaluated as a supplemental analysis.

Stata 14.1 was used to calculate accuracy parameters and SROC parameters. Review manager 5.3 was employed to construct forest plots and SROC graphs.

Investigations of heterogeneity

As suggested in Cochrane handbook for systematic reviews of diagnostic test accuracy, the confidence region depicts uncertainty in the overall average value caused by sampling variability, the prediction region depicts variation from between study heterogeneity. Where heterogeneity is high, review authors will note that the 95% prediction region is much larger than the 95% confidence region. Prediction regions also take account of correlations in variation in sensitivity and specificity, and variation in positivity threshold. No equivalent to the I2 statistic is currently available for diagnostic test accuracy meta-analysis. Computing separate I2 statistics for sensitivity and specificity fails to account for variation explained by threshold effects, and the correlation of sensitivity and specificity, and will overestimate the degree of heterogeneity observed [30].

Stata 14.1 was used for computing the 95% confidence and 95% prediction regions.

Results

A total of 3834 potentially relevant articles were initially found by searching the electronic databases and other sources. After duplicate removal and exclusion of articles judged irrelevant based on the screening of titles and abstracts, 54 records were reviewed as full texts. Of these, 37 articles met the eligibility criteria and were included for qualitative analysis [5–21,31–50].

Fig. 1 shows the flowchart of the study selection process.

Qualitative analysis

Description of studies

The characteristics of the 37 studies included in the qualitative analysis are shown in Table 1. Table S2 presents the list of the studies judged not eligible for the qualitative analysis with reasons for their exclusion.

The 37 articles included participants from different countries: Canada (n = 4), USA (n = 14), Spain (n = 1), Germany (n = 1), Greece (n = 1), Netherlands (n = 1), UK (n = 1), Turkey (n = 1), Australia (n = 2), Sweden (n = 1), China (n = 1), Hong Kong (n = 2), Taiwan (n = 2), Chile (n = 1), Brazil (n = 2) and Thailand (n = 2).

Most of the studies were performed on children and adolescents with sleep disordered breathing symptoms or on a general pediatric population; a variety of other conditions were also included,

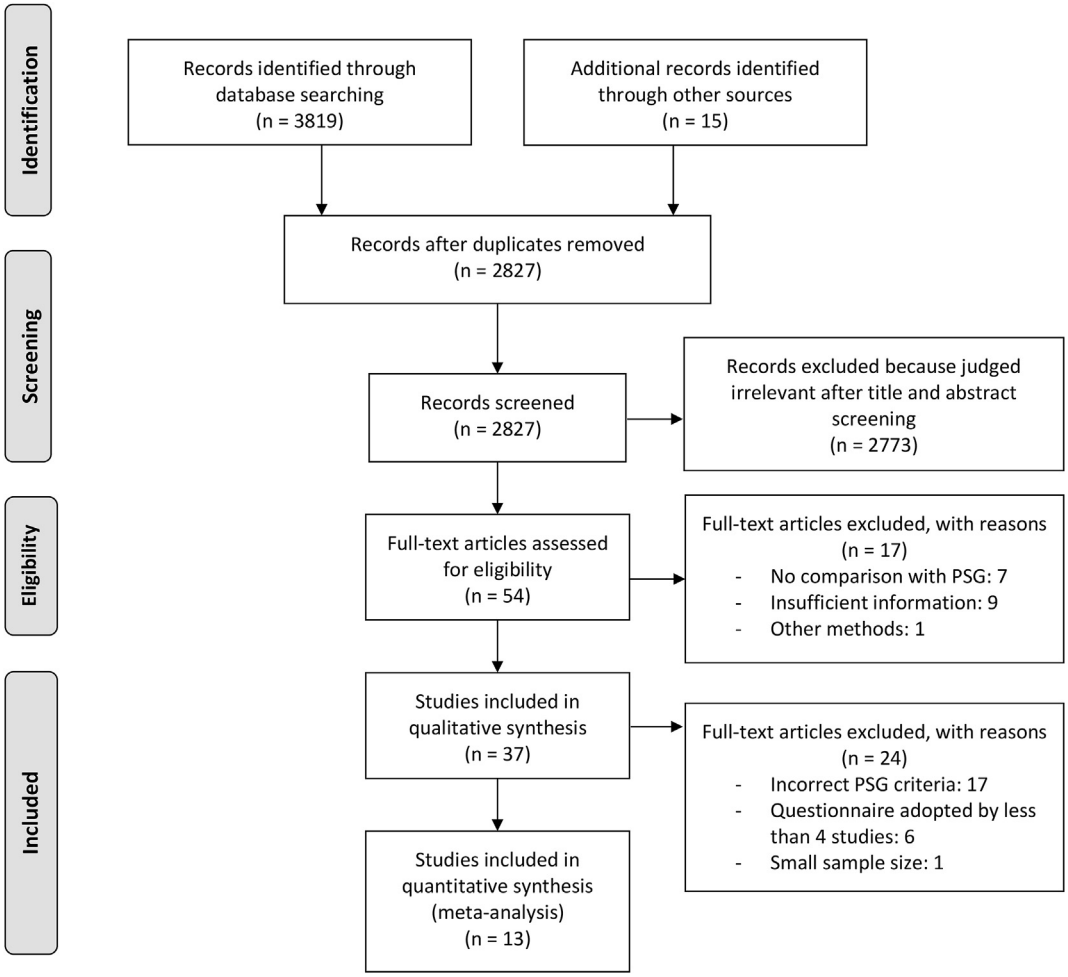

Fig. 1. Flowchart of the literature search.

Table 1
Description of studies included in the qualitative analysis.

N	First author, year of publication	Questionnaire	Country	Age (years)	Population	Sample size	PSG criteria
1	Abumuamar et al., 2018 [34]	IF SLEEPY	Canada	2–18	Children referred for a sleep evaluation	326	AHI ≥ 1.5 AHI ≥ 5
2	Ahmed et al., 2018 [6]	IMP-QESS-CHAD OSA-18 PSQ	USA	5–9.9	Children with OSA	86	AHI ≥ 5 AHI ≥ 10
3	Alonso-Álvarez et al., 2014 [19]	Abbreviated PSQ (Spanish validated version)	Spain	3–14	Obese children	248	RDI ≥ 3
4	Amin et al., 2015 [14]	PSQ Pediatric modification of ESS	Canada	0–18	Children with severe chronic kidney failure	19	OAHI > 2
5	Bertran et al., 2015 [31]	PSQ (Spanish validated version) PSQ-6	Chile	0–15	Habitually snoring children referred for a sleep evaluation	83	AHI ≥ 1
6	Bhushan et al., 2014 [35]	14-ItemScore	USA	0–18	Children with OSA	97	AHI ≥ 1
7	Biggs et al., 2014 [36]	PSSI	Australia	5–18	Children referred for a sleep evaluation	142	AHI > 5
8	Borgstrom et al., 2013 [32]	OSA-18	Sweden	1–12	Children having undergone PSG	225	AHI > 1 AHI ≥ 5 AHI ≥ 10
9	Carno et al., 2008 [5]	PSQ	USA	8–17	Overweight children with habitual snoring referred for sleep evaluation	96	AHI ≥ 5
10	Carroll et al., 1995 [37]	Expanded OSA score	USA	5–15	Children with sleep-disordered breathing	83	AHI ≥ 1
11	Chan et al., 2009 [7]	Modified ESS (Chinese validated version)	China	3–12	Children with suspected OSA	192	AHI > 5
12	Chan et al., 2012 [12]	Taiwan Chinese version of SRBD scale	Hong Kong	2–18	Children with suspected OSA	102	AHI > 1.5
13	Chervin et al., 2007 [21]	The SRDB scale of the PSQ	USA	5–13	Children scheduled for adenotonsillectomy	105	OAI ≥ 1
14	Cielo et al., 2014 [15]	PSQ	USA	2–18	Children with craniofacial disorders who performed PSG and completed PSQ	83	AHI > 2 AHI > 5
15	Chervin et al., 2000 [8]	PSQ	USA	2–18	Children with PSG confirmed SRBD and general pediatric subjects	162	AHI > 5
16	Ehsan et al., 2017 [20]	PSQ	USA	2–18	Children with asthma referred for PSG	160	OI > 1 OI > 2 OI > 5
17	Ferreira et al., 2009 [38]	SDSC	Brazil	3–17	Various	40	AHI > 1
18	Goodwin et al., 2004 [39]	TuCASA-SHQ	USA	6–11	Various	480	RDI > 1
19	Ishman et al., 2015 [40]	OSA-18	USA	2–12	Children with OSA	79	OAHI > 1 OAHI ≥ 5 OAHI ≥ 10
20	Ishman et al., 2016 [41]	PSQ	USA	0–18	Obese children at risk of OSA	45	OAHI ≥ 1 OAHI ≥ 5
21	Kadmon et al., 2013 [9]	PosaST	Canada	3–9	Children referred for sleep evaluation	85	AHI ≥ 1.5 AHI ≥ 5
22	Kadmon et al., 2014 [13]	IF SLEEPY Pediatric modified STOP-Bang	Canada	3–18	Children referred for sleep evaluation	137	AHI ≥ 1.5
23	Kaewkul et al., 2018 [18]	Modified OSA-18 (Thai validated version)	Thailand	2–12	Snoring children who performed PSG	123	AHI ≥ 10
24	Kang et al., 2012 [42]	OSA-18 (Chinese validated version)	Taiwan	2–18	Children with sleep problems	109	AHI ≥ 1
25	Li et al., 2006 [43]	HK-CSQ (Chinese version)	Hong Kong	5–15	Children with suspected OSA	229	OAI ≥ 1
26	Longlalerng et al., 2018 [17]	Thai SRDB-PSQ (validation)	Thailand	7–18	Overweight or obese children	62	AHI ≥ 1 AHI ≥ 4
27	Mousailidis et al., 2014 [44]	Greek OSA-18 (validation)	Greece	3–18	Snoring children referred for PSG	141	AHI ≥ 1
28	Owens et al., 2000 [45]	CSHQ	USA	5–9	Children with sleep problems	623	RDI > 1
29	Pires et al., 2017 [10]	Brazilian Portuguese PosaST	Brazil	3–9	Children referred for PSG	60	AHI ≥ 5
30	Plomp et al., 2012 [46]	Brouillette questionnaire	Netherlands	<17 y	Children with Treacher-Collins Syndrome	13	AHI > 1
31	Schnoor et al., 2018 [11]	PSQ-SRDB subscale OSAsq8 OSA3/8	Germany	<18 y	Children referred for PSG for suspected OSA	53	AHI > 5
32	Sproson et al., 2009 [47]	PSQ	UK	3–8	Snoring children listed for adenotonsillectomy, tonsillectomy, adenoidectomy	58	AHI ≥ 1 AHI ≥ 5
33	Spruyt et al., 2012 [16]	PosaST 37 items Sleep Questionnaire	USA	5–9	Various	1133	1 < AHI ≤ 2 2 < AHI ≤ 3 3 < AHI ≤ 5 5 > AHI < 10 AHI ≥ 10
34	Uyan et al., 2016 [48]	PSQ (Turkish validated version)	Turkey	3–18	Children with bronchiolitis obliterans	21	OAHI > 1

Table 1 (*continued*)

N	First author, year of publication	Questionnaire	Country	Age (years)	Population	Sample size	PSG criteria
35	Walter et al., 2015 [33]	OSA-18	Australia	6 mo-16.4	Children referred for sleep evaluation versus non snoring children	216	OAHI > 2 OAHI > 5
36	Ward et al., 2017 [49]	PSQ	USA	6–11	Children with juvenile idiopathic arthritis	143	OAHI ≥ 1.5
37	Yang et al., 2010 [50]	Chinese version of the PDSS	Taiwan	11–15	Patients recruited from a sleep disorder center	62	AHI > 1

AHI, apnea hypopnea index; AI, apnea index; CSHQ, children's sleep habits questionnaire; ESS-CHAD, Epworth sleepiness scale for children and adolescents; HK-CSQ, Hong Kong children sleep questionnaire; IMP-Q, important questions questionnaire; OAHI, obstructive apnea hypopnea index; OAI, obstructive apnea index; OI, obstructive index; OSA, obstructive sleep apnea; PDSS, pediatric daytime sleepiness scale; PosaST, pediatric obstructive sleep apnea screening tool; PSG, polysomnography; PSQ, pediatric sleep questionnaire; PSSI, pediatric sleep survey instrument; RDI, respiratory disturbance index; SDB, sleep disordered breathing; SDSC, sleep disturbance scale for children; SHQ, sleep habits questionnaire; TuCASA-SHQ, Tucson children's assessment of sleep apnea-sleep habits questionnaire.

such as obesity (n = 4), kidney problems (n = 1), craniofacial disorders (n = 1), asthma (n = 1), chronic lung disease (n = 1), juvenile idiopathic arthritis (n = 1), Treacher-Collins syndrome (n = 1), and children undergoing adenotonsillectomy (n = 2). Sample size varied widely, ranging from 13 to 1133 participants.

Twenty different screening questionnaires were identified from the 37 studies included in the qualitative analysis. The list of the translated versions available for each of these questionnaires was shown in Table S3.

Quality assessment

No studies were judged at high risk for bias. Therefore, no studies were excluded after quality assessment. Description of blinding to results of the PSG when analyzing questionnaires was frequently missing. Another frequent limitation was the small sample size of many studies and the lack of a sample size calculation. The results of methodological quality of studies included in the qualitative analysis are shown in Fig. 2.

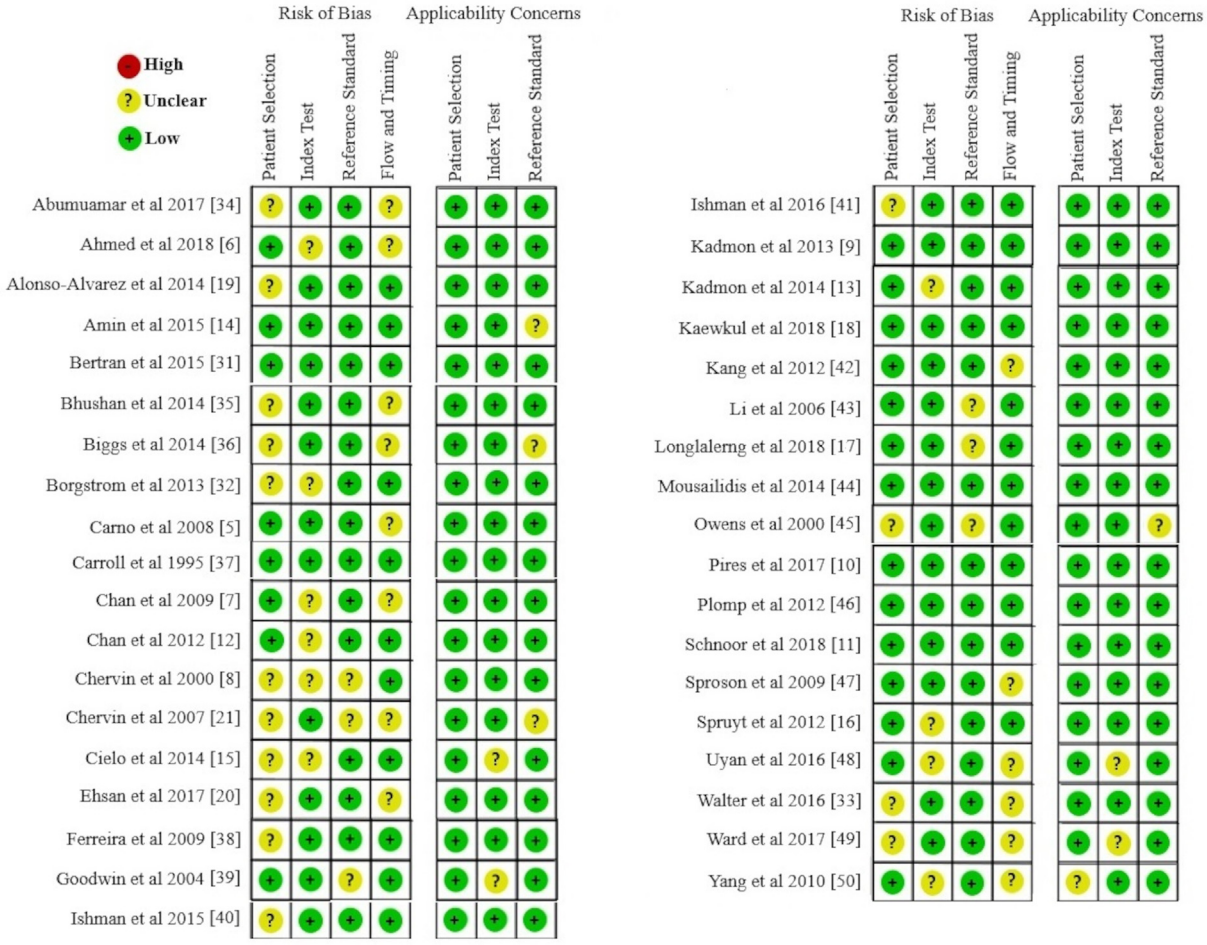

Fig. 2. Risk of bias and applicability concerns summary using the QUADAS-2 (quality assessment of diagnostic accuracy studies) tool. Key domains: patient selection; index test; reference standard; study flow and timing.

Quantitative analysis

Data synthesis and findings

Among the 37 articles included in the qualitative analysis, 13 met the eligibility criteria required for quantitative analysis and were included in further analyses (Fig. 1).

The sleep-related breathing disorder scale of the pediatric sleep questionnaire (SRBD-PSQ) and the OSA-18 were the only questionnaires adopted by at least four studies with AHI≥1 being used as diagnostic threshold and, therefore, were included in the quantitative analysis. For these questionnaires, the studies with AHI≥5 as cutoff were also evaluated as supplemental analysis (SRBD-PSQ: four studies for AHI≥1 and seven for AHI≥5; OSA-18: four studies for AHI≥1 and four for AHI≥5). Description of studies with AHI≥1 and AHI≥5 as diagnostic threshold is showed, respectively, in Table 2 and Table 3. The list of studies judged not eligible for quantitative analysis with reasons for their exclusion is shown in supplementary data (Table S4).

Using AHI≥1 as diagnostic cutoff (mild OSA), SRBD-PSQ was more sensitive (0.76 [95% confidence interval (CI) 0.65, 0.84]) than OSA-18 (0.56 [95% CI 0.51, 0.61]), while OSA-18 resulted to have a higher specificity (0.73 [95% CI 0.53, 0.87]) compared to SRBD-PSQ (0.43 [95% CI 0.27, 0.60]). Focusing on SRBD-PSQ studies defining OSA with an AHI≥5 (moderate-severe OSA), this tool exhibited slightly higher accuracy parameters (sensitivity 0.79 [95% CI 0.69, 0.86]; specificity 0.47 [95% CI 0.28, 0.67]). The overall results are shown in Table 4.

Forest plots and SROC curves of studies with AHI≥1 as diagnostic threshold are shown in Figs. 3–6. Forest plots and SROC curves of studies with AHI≥5 as diagnostic threshold are shown in supplementary data (Figures S1–S4).

A short version of the PSQ (PSQ-6) was employed in a study included in the quantitative analysis [31]; it was not included in the overall results calculations, but it has been taken into consideration for completeness. For PSQ-6, using AHI≥1 as cutoff, sensitivity resulted 0.89 and specificity 0.54.

Table 2
Description of studies included in the quantitative analysis with AHI≥1 as polysomnographic cutoff.

Study	Questionnaire	PR	SE	SP	PPV	NPV	LR+	LR-	DOR	TP	FP	FN	TN
Bertran et al., 2015 [31]	PSQ 6-item	42.2	88.6	53.8	53.4	84	1.57	0.26	9.05	31	22	4	26
Bertran et al., 2015 [31]	PSQ	42.2	71.4	52.1	52.1	71.4	1.49	0.55	2.72	25	23	10	25
Borgstrom et al., 2013 [32]	OSA-18	90.22	55.2	40.9	89.6	9	0.93	1.1	0.85	112	13	91	9
Ishman et al., 2015 [40]	OSA-18	89.9	54.9	75	95.1	15.8	2.2	0.6	3.65	39	2	32	6
Ishman et al., 2016 [41]	PSQ	84.4	82	71	94	42	2.83	0.25	11.15	31	2	7	5
Kang et al., 2012 [42]	OSA-18	71.56	62.8	83.9	90.7	47.3	3.9	0.44	8.8	49	5	29	26
Longlalerng et al., 2018 [17]	PSQ	59.7	62	60	69.66	51.59	1.55	0.63	2.45	23	10	14	15
Mousailidis et al., 2014 [44]	OSA-18	62.4	53.4	83	83.9	51.77	3.14	0.56	5.59	47	9	41	44
Sproson et al., 2009 [47]	PSQ	79.69	84.8	16.6	79.96	21.77	1.02	0.92	1.11	39	10	7	2

AHI, apnea hypopnea index; DOR, diagnostic odd ratio; FN, false negative; FP, false positive; LR-, negative likelihood ratio; LR+, positive likelihood ratio; NPV, negative predictive value (%); OSA, obstructive sleep apnea; PPV, positive predictive value (%); PR, prevalence (%); PSQ, pediatric sleep questionnaire; SE, sensitivity (%); SP, specificity (%); TN, true negative; TP, true positive.

Table 3
Description of studies included in the quantitative analysis with AHI≥5 as polysomnographic cutoff.

Study	Questionnaire	PR	SE	SP	PPV	NPV	LR+	LR-	DOR	TP	FP	FN	TN
Ahmed et al., 2018 [6]	PSQ	51.16	72.73	61.9	66.67	68.42	1.91	0.44	4.33	16	8	13	6
Ahmed et al., 2018 [6]	OSA-18	51.16	59.09	57.14	59.09	57.14	1.38	0.72	1.93	13	9	12	9
Borgstrom et al., 2013 [32]	OSA-18	56.88	59.3	48.5	60.30	47.50	1.15	0.84	1,37	76	50	52	47
Carno et al., 2008 [5]	PSQ	61.5	84	23	63.54	47.37	1.09	0.70	1.57	50	28	9	9
Chervin et al. (A) 2000 [8]	PSQ	85	85	87	97.37	50.58	6.54	0.17	37.92	59	2	10	11
Chervin et al. (B) 2000 [8]	PSQ	85	81	87	97.25	44.69	6.23	0.22	28.53	56	2	13	11
Cielo et al., 2014 [15]	PSQ	27.7	57	48	30.00	74.00	1.10	0.90	1.22	13	31	10	29
Ishman et al., 2015 [40]	OSA-18	59.5	56.3	54.8	65.9	44.7	1.25	0.80	1.56	26	14	21	18
Ishman et al., 2016 [41]	PSQ	48.9	86	38	55.00	75.00	1.39	0.37	3.76	19	14	3	9
Sproson et al., 2009 [47]	PSQ	31.14	88.2	17.1	32.48	76.22	1.06	0.69	1.54	16	33	2	7
Walter et al., 2015 [33]	OSA-18	21.3	93	25	25.13	92.96	1.24	0.28	4.43	43	127	3	42

AHI, apnea hypopnea index; DOR, diagnostic odd ratio; FN, false negative; FP, false positive; LR-, negative likelihood ratio; LR+, positive likelihood ratio; NPV, negative predictive value (%); OSA, obstructive sleep apnea; PPV, positive predictive value (%); PR, prevalence (%); PSQ, pediatric sleep questionnaire; SE, sensitivity (%); SP, specificity (%); TN, true negative; TP, true positive.

Table 4
Description of summary estimate for SRBD-PSQ and OSA-18.

	PSQ AHI ≥ 1 Summary estimate (95% CI)	PSQ AHI ≥ 5 Summary estimate (95% CI)	OSA-18 AHI ≥ 1 Summary estimate (95% CI)	OSA-18 AHI ≥ 5 Summary estimate (95% CI)
Number of studies	4	7	4	4
Number of participants	248	487	554	563
Sensitivity	0.76 (0.65 0.84)	0.79 (0.69 0.86)	0.56 (0.51 0.61)	0.69 (0.45 0.85)
Specificity	0.43 (0.27 0.60)	0.47 (0.28 0.67)	0.73 (0.53 0.87)	0.44 (0.31 0.58)
Diagnostic odds ratio	2.34 (1.24 4.43)	3.32 (1.24 8.86)	3.59 (1.38 9.32)	1.70 (0.94 3.07)
Positive likelihood ratio	1.33 (1.02 1.73)	1.49 (0.99 2.24)	2.13 (1.07 4.20)	1.22 (1.03 1.44)
Negative likelihood ratio	0.57 (0.38 0.84)	0.45 (0.25 0.82)	0.59 (0.45 0.78)	0.72 (0.46 1.12)

AHI, apnea–hypopnea index; OSA, obstructive sleep apnea; SRBD-PSQ, sleep-related breathing disorder scale of the pediatric sleep questionnaire.

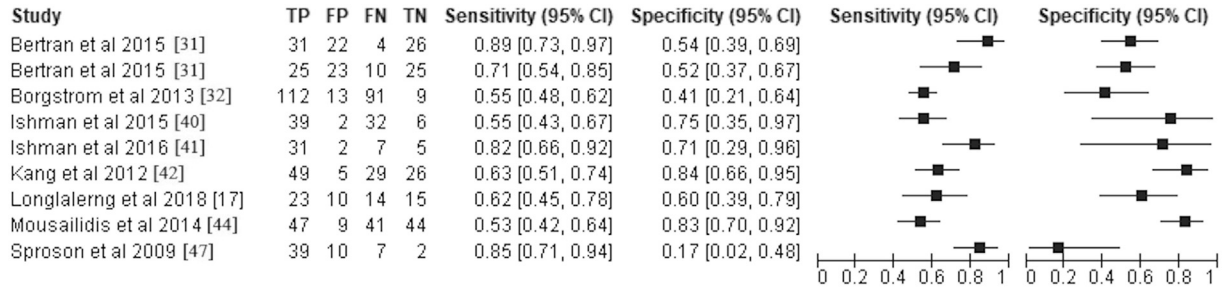

Fig. 3. Forest plot of all studies with AHI≥1 as polysomnographic cutoff. AHI, apnea–hypopnea index; CI, confidence interval; FN, false negative; FP, false positive; TN, true negative; TP, true positive.

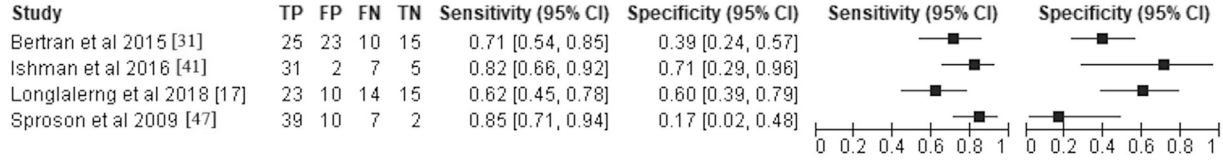

Fig. 4. Forest plot of SRBD-PSQ studies with AHI≥1 as polysomnographic cutoff. AHI, apnea–hypopnea index; CI, confidence interval; FN, false negative; FP, false positive; TN, true negative; TP, true positive.

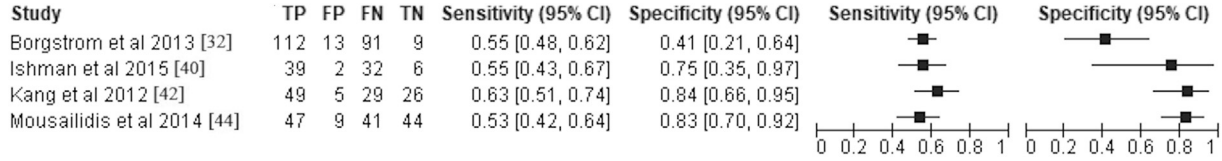

Fig. 5. Forest plot of OSA-18 studies with AHI≥1 as polysomnographic cutoff. AHI, apnea–hypopnea index; CI, confidence interval; FN, false negative; FP, false positive; TN, true negative; TP, true positive.

Heterogeneity of included studies

The extensive surfaces of the prediction regions of SROC curves (Fig. 6) were related to the significant heterogeneity of the included studies. It was explained by the different population conditions of the studies of this review (obesity, snoring, undergoing adenotonsillectomy, OSA, craniofacial disorders, sleep disordered breathing, sleep problems, various). Another explanation could be the large OSA prevalence range in studies populations (between 21.3 and 90.22%) and the large age range (between 1 and 18 y).

Discussion

Well-conducted systematic reviews regarding a disease at high-risk of medical consequences in the growing patient, such as OSA, represent valuable resources for both researchers and clinicians,

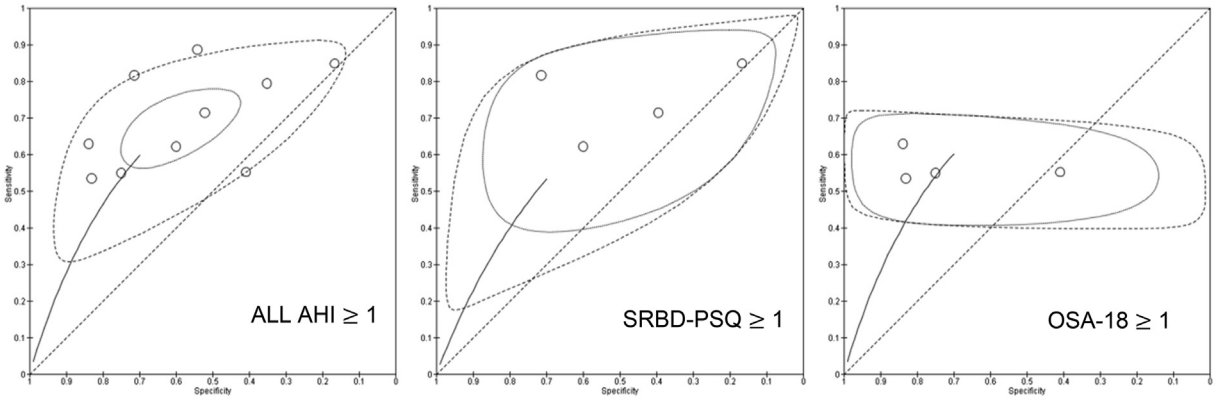

Fig. 6. SROC curve, 95% confidence region (full line area) and 95% prediction regions (dashed line area) of studies with AHI≥1 as polysomnographic cutoff. All studies; SRBD-PSQ studies; OSA-18 studies. AHI, apnea–hypopnea index; OSA, obstructive sleep apnea; SRBD-PSQ, sleep-related breathing disorder scale of the pediatric sleep questionnaire; SROC, summary receiver operator characteristic.

summarizing the relevant available findings and enabling rational decision-making. There is significant appeal in the concept of a practical and simple tool that can assist in the screening of pediatric OSA, potentially aid the referral process to sleep medicine specialists and the prioritization of the long wait lists at specialists for children already referred. Questionnaires should be particularly relevant because the current confirmatory procedure for OSA is cost-prohibitive and mainly available in specialized sleep centers. Diagnostic test accuracy reviews play an important role in order to identify where sufficient evidence exists for or against the use of a screening questionnaire for pediatric OSA but, in order to ensure clinical applicability and findings generalizability, it is necessary to compare studies with the same reference standard parameter in order to avoid a significant source of bias [51]. This systematic review and meta-analysis investigated the characteristics of available questionnaires using the currently accepted diagnostic threshold for pediatric OSA (AHI $\geq$1), and was conducted according to the guidelines of the Cochrane handbook for systematic reviews of diagnostic test accuracy.

Screening tools for OSA should be selected according to their sensitivity rather than their specificity because, through the detection of more true positives, they should facilitate early diagnosis of pediatric OSA and aid the referral process for PSG examination [24].

A recent diagnostic meta-analysis of Wu et al. [24] recommended the combined use of SRBD-PSQ and pulse oximetry for early detection of pediatric OSA when PSG is not available [24]. According to the reviews of Michelet et al. [22] and Canto et al. [25], the SRBD-PSQ had a diagnostic accuracy good enough to be used as a screening method. Accordingly, SRBD-PSQ proved to be the most sensitive questionnaire in the present study and, if clinicians want to use a screening questionnaire, this showed the most robust characteristics. Sensitivity and specificity found in our review were different from those detected in the original study performed by Chervin et al. [8] (respectively, 0.76 *versus* 0.81 and 0.43 *versus* 0.87). The heterogeneous population included in this meta-analysis, as demonstrated by the extensive surface of the prediction region in SROC curves, can explain these controversial findings. Another reason could be the difference between the polysomnographic cutoff parameter used in this review to define the presence of OSA (AHI$\geq$1) and the one adopted by Chervin et al. [8] (AHI$\geq$5). Several short-form versions of screening questionnaires for pediatric OSA have been developed. In the present study PSQ-6 [31] has been considered, with slightly improved accuracy parameters being found (sensitivity of 0.89 and specificity of 0.54).

OSA-18 questionnaire was originally developed as a specific quality of life tool, with a poor validity in determining the presence of OSA [32,33]. Accordingly, our results confirmed a poor sensitivity for this questionnaire (0.56).

This systematic review has some limitations that should be acknowledged. Study populations may differ for many characteristics such as age, health condition and ethnicity. Most of the studies included populations from sleep centers or children with snoring or breathing symptoms, a fact that can affect the prevalence of OSA in the sample and, also, reduce applicability of the results to the general pediatric population. Most articles had no blinding interpretation of the reference and index tests; the index test may be biased if the assessor knows PSG results and, similarly, the analysis of PSG should be conducted blinded to the questionnaire results. Finally, the time interval between the index test and the PSG was generally not mentioned. Another limitation is the small number of studies available for quantitative analysis. However, no high risk of bias was observed using the QUADAS-2 tool.

Conclusions

Screening questionnaires for OSA should be particularly useful when needing to determine the presence of OSA because PSG, as the current confirmatory procedure, is cost-prohibitive and not widely available. This systematic review identified two screening questionnaires with acceptable characteristics. Of these, SRBD-PSQ performed best and showed the highest sensitivity using the currently accepted diagnostic threshold for pediatric OSA (AHI $\geq$1). However, there is still insufficient evidence to replace PSG as the current reference standard for the diagnosis of OSA in children. Further well-designed studies are still warranted to investigate the role of SRBD-PSQ in real-world clinical populations.

Practice points

1. The sleep-related breathing disorder scale of the pediatric sleep questionnaire (SRBD-PSQ) is a sensitive tool for detecting pediatric obstructive sleep apnea (OSA).
2. The SRBD-PSQ yields slightly better sensitivity and specificity in screening moderate pediatric OSA.
3. Further well-designed studies are still warranted to investigate the role of SRBD-PSQ in real-world clinical populations in order to assist clinicians in the early identification of possible OSA in children, thus aiding the referral process to sleep specialists or being used as a triage screening tool for children already referred.

Research agenda

1. None of the questionnaires investigated in this systematic review and meta-analysis exhibited optimal accuracy for the diagnosis of pediatric OSA.
2. Further well-conducted studies for the identification of more accurate screening tools should be encouraged in order to reduce underdiagnosis of OSA among children.
3. Studies on the accuracy of screening questionnaire should not adopt apnea–hypopnea index (AHI) thresholds different from the established value for classifying the presence of pediatric OSA in order to reduce a potentially significant source of bias in both primary and secondary research.

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Conflicts of interest

The authors do not have any conflicts of interest to disclose.

Appendix A. Supplementary data

Supplementary data to this article can be found online at https://doi.org/10.1016/j.smrv.2021.101464.

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