Received: 13 April 2018 | Revised: 5 November 2018 | Accepted: 18 December 2018

DOI: 10.1111/jsr.12826

REGULAR RESEARCH PAPER

Journal of Sleep Research

Screening for sleep-disordered breathing with Pediatric Sleep Questionnaire in children with underlying conditions

Rishi Pabary[1,2*] | Christophe Goubau[1,3*] | Kylie Russo[1] | Aidan Laverty[1] | François Abel[1] | Martin Samuels[1]

[1]Department of Paediatric Respiratory Medicine, Great Ormond Street Hospital for Children, London, UK

[2]Department of Paediatric Respiratory Medicine, Royal Brompton and Harefield NHS Foundation Trust, London, UK

[3]Department of Paediatric Respiratory Medicine, Cliniques Universitaires Saint Luc, Brussels, Belgium

Correspondence
Martin Samuels, Respiratory Unit, Great Ormond Street Hospital for Children NHS Foundation Trust, London, UK.
Email: Martin.samuels@gosh.nhs.uk

Abstract

The Pediatric Sleep Questionnaire described by Chervin et al. (Sleep Medicine, 2000, 1, 21–32) was originally validated for children with obstructive sleep apnoea syndrome but without other disorders. The aim of our study was to check the applicability of this questionnaire in children with underlying chronic medical conditions. Children aged 2–18 years who underwent a diagnostic sleep study at Great Ormond Street Hospital were recruited over a 10-month period. The Pediatric Sleep Questionnaire completed by their parents and cardiorespiratory polygraphy were scored. Sensitivities and specificities of the Pediatric Sleep Questionnaire were calculated using a Pediatric Sleep Questionnaire score of 0.33 as being indicative of sleep-disordered breathing. A total of 561 patients were reviewed. Neuromuscular disorders (n = 108), craniofacial anomalies (n = 58) and the obstructive sleep apnea syndrome control group (n = 155) were best represented. The sensitivity for patients with isolated obstructive sleep apnoea syndrome was 76.5% when using an apnoea–hypopnoea index ≥ 5, but this was much lower when looking at specific sub-groups such as neuromuscular patients (25%) or patients with Trisomy 21 (36.7%). Sensitivities remained unchanged for patients with obstructive sleep apnoea syndrome (77.3%) when an apnoea–hypopnoea index of ≥ 1 was used, but improved for neuromuscular disorders sub-groups (36.7%) and Trisomy 21 (84%). In conclusion, the Pediatric Sleep Questionnaire is not a good screening tool for obstructive sleep apnoea syndrome in children with complex underlying disorders when a cut-off apnoea–hypopnoea index of ≥ 5 is used, and it cannot replace cardiorespiratory polygraphy recording.

KEYWORDS

craniofacial, neuromuscular, paediatrics, sleep apnea syndromes, surveys and questionnaires, Trisomy 21

1 | INTRODUCTION

Obstructive sleep apnoea syndrome (OSAS) is common in children, with a prevalence ranging from 1.2% to 5.8%, depending on the population studied and the defining criteria (Bixler et al., 2009; Li et al., 2010; Lumeng & Chervin, 2008). Sleep-disordered breathing (SDB), including OSAS, is acknowledged to be an important cause of morbidity in children, with behavioural and neurocognitive abnormalities more common in children with SDB than in those without SDB (Galland et al., 2015; Marcus et al., 2012;

*R. P. and C. G. are joint first authors.

Schechter, 2002). A number of chronic medical conditions exist in which SDB is common and consequent morbidity more likely, including Trisomy 21 (T21; Maris, Verhulst, Wojciechowski, Van de Heyning, & Boudewyns, 2016), neuromuscular disease (Arens & Muzumdar, 2010), sickle cell disease (Whitesell et al., 2016) and craniofacial abnormalities (Cielo et al., 2014; MacLean, Fitzsimons, Hayward, Waters, & Fitzgerald, 2008; RCPCH Working Party on Sleep Physiology, 2009).

The gold-standard for the diagnosis of SDB is polysomnography (PSG; AAP, 2002; Brouilette et al., 1984; Marcus et al., 2012); this requires time, is resource intensive and requires highly-trained sleep physiologists to perform accurately. To address this issue, paediatric predictive tools for SDB have been designed to select those children that warrant further investigation with PSG. The Pediatric Sleep Questionnaire (PSQ) developed by Chervin, Hedger, Dillon, and Pituch (2000) was prospectively validated as a reliable screening tool for SDB (Chervin et al., 2000). It was demonstrated to have a sensitivity of 85% and specificity of 87% for identifying SDB confirmed by PSG in otherwise healthy children aged 2–18 years. SDB was defined as a value of apnoea–hypopnoea index (AHI) ≥ 5, a level where surgical intervention is commonly recommended. Although this PSQ was originally validated for children with OSAS aged 2–18 years after exclusion of those with severe medical or mental impairments, it has since been used in many paediatric populations in which the questionnaire was not validated, including in children with somnambulism (Guilleminault et al., 2005), autism (Clarke et al., 2005), high or low body mass index (BMI; Vaher, Kasenõmm, Vasar, & Veldi, 2013), children with nocturnal enuresis (Wolfe-Christensen, Kovacevic, Mirkovic, & Lakshmanan, 2013) and asthma (Ehsan, Kercsmar, Collins, & Simakajornboon, 2017). The applicability of the PSQ in children who have other underlying chronic medical conditions at risk for SDB, including T21, neuromuscular disease, sickle cell or craniofacial abnormalities, remains unclear. In this paper, we aimed to evaluate the relationship between the Chervin PSQ score and cardiorespiratory polygraphy (CRP) findings to determine the utility of the PSQ in detecting OSAS in this group of children with chronic underlying conditions. We hypothesized that the PSQ is a valid tool in this group.

2 | METHODS

2.1 | Subjects

Children aged 2–19 years who underwent a diagnostic sleep study (CRP) in the Paediatric Sleep Unit at Great Ormond Street Hospital (GOSH), London, for clinical indications including symptoms suggestive of SDB and patients at risk for SDB, were recruited over a 10-month period retrospectively (October 2015 to January 2016) and prospectively (between February 2016 to August 2016). Data were assessed from chart review. Patients were categorised according to clinical diagnosis, with no overlap between patients' sub-groups. The PSQ described by Chervin et al. (2000) was given to parents of patients as part of routine clinical care on the night of the sleep study. We excluded children if oxygen or non-invasive ventilation (NIV) settings were titrated during the overnight CRP as this was likely to impact on the AHI, oxygen desaturation index (ODI) and transcutaneous carbon dioxide ($TcPCO_2$) levels over the duration of the study.

2.2 | Pediatric Sleep Questionnaire

The PSQ scores 22 items that ask about snoring frequency, loud snoring, observed apnoeas, difficulty breathing during sleep, daytime sleepiness, inattentive or hyperactive behaviour, and other paediatric OSAS features, each previously shown to correlate with OSAS confirmed by PSG in referred children (Chervin et al., 2000). The items are divided into nocturnal, daytime and cognitive symptoms. Each item is scored as being present, absent or unknown. The score was calculated by dividing the number of symptoms that are present by the total number of symptoms that are present or absent (present/present + absent); any questions that are not answered (unknown) are not included in the calculation. The score can vary from 0 to 1. Previous data suggest that a cut-off value of 0.33 was most effective in identifying paediatric OSAS (Chervin et al., 2000, 2007; Clarke et al., 2005; Ehsan et al., 2017; Guilleminault et al., 2005; Vaher et al., 2013; Wolfe-Christensen et al., 2013). Paper copies of the PSQ were completed by parents of patients admitted for CRP.

2.3 | Cardiorespiratory polygraphy

A nocturnal CRP was performed and scored using adapted rules as per the 2012 American Academy of Sleep Medicine (AASM) guidelines for the scoring of sleep and associated events (Berry & Gamaldo, 2012). The studies included nasal pressure transducer, thoracic and abdominal excursion via respiratory inductance plethysmography, 2-lead electrocardiography, pulse oximetry (Nonin via Natus Embla recording device supplied by Stowood Scientific Instruments, UK), transcutaneous pCO_2 (Tosca 500, Radiometer, Denmark) and infra-red video recording with microphone. The digital equipment used was Embla S4500 (Stowood Scientific Instruments, UK). This abbreviated CRP without electroencephalographic, electrooculographic or electromyographic leads has previously been demonstrated to be an accurate tool for the detection of OSAS (Alonso Álvarez et al., 2008; Jacob et al., 1995). Sleep stages were scored as either wake, rapid eye movement or non-rapid eye movement sleep in 30-second epochs by visual analysis of the cardiorespiratory parameters (Redmond et al., 2007; Vaughn, Quint, Messenheimer, & Robertson, 1995).

An obstructive apnoea was scored if there was > 90% fall in nasal pressure transducer for > 90% of the entire event, the event lasted ≥ 2 breaths, and there was continued or increased respiratory effort. A mixed apnoea was scored when there was absent respiratory effort during one portion of the event and the presence

of inspiratory effort in another portion, regardless of which portion comes first. A central apnoea was scored if there was absent respiratory effort for the entire event, and either the event lasted for > 20 s or lasted ≥ 2 breaths and was associated with a ≥ 3% oxygen desaturation. A hypopnoea was scored if there was a ≥ 30% reduction in amplitude of the nasal pressure transducer, the event lasted for ≥ 2 breaths and was associated with a ≥ 3% oxygen desaturation. The final AHI was determined by dividing the total number of apnoeas and hypopnoeas by the number of hours of total sleep time. A value of AHI of ≥ 5 or a PSQ score > 0.33 was indicative of SDB (Chervin et al., 2000). Obstructive AHI (oAHI) was determined by dividing the total number of obstructive apnoeas, mixed apnoeas and hypopnoeas by the number of hours of total sleep time. The ODI (defined as the number of dips in oxygen saturation ≥ 3% per hour of sleep time) was also calculated during study analysis.

2.4 | Data analysis

Patients tested were divided according to their underlying disease; for an individual disease group to have a separate classification at least 20 sleep studies were needed in each group. Primary diagnoses were considered for classification. The resulting categories were "neuromuscular", "craniofacial", "T21", "haemoglobinopathies" (Hb), "OSAS" and "other." Sensitivity, specificity, positive predictive value (PPV) and negative predictive value (NPV) of the PSQ were calculated for the different disease groups. The differences between groups were analysed with non-parametric Kruskal–Wallis with Dunn's multiple comparison test. We set the level of statistical significance at .05. Analyses were performed using a statistical software program (GraphPad Prism 6.0; GraphPad Software, San Diego, CA, USA). Summary measures are given as medians with interquartile range.

In the original paper validating the use of this PSQ, AHI ≥ 5 was taken as indicative of OSAS. However, in another paediatric study of normal children, an AHI ≥ 1 was considered abnormal (Marcus, 1992). All statistical analyses were therefore repeated using this cut-off value to see whether this had any impact on the sensitivity, specificity, PPV and NPV of the PSQ.

Correlations between PSQ score, oAHI and total AHI were also calculated using non-parametric Spearman Rank Correlation; none of the variables passed statistical analyses of normal distribution.

Children with chronic diseases can have a cognitive impairment as part of their underlying disease that can affect parental interpretation of the cognitive domain of the PSQ (Masters, Harvey, Wales, O'Callaghan, & Harris, 1999). We therefore assessed, in a sub-group analysis, the sensitivity and specificity of the PSQ score with and without the cognitive domain included in the analysis for children referred with simple OSA (n = 69) and for those with other underlying medical conditions (n = 230). The data collection to address this part of the study question was commenced prospectively in April 2016.

3 | RESULTS

3.1 | Patients' characteristics

Pediatric Sleep Questionnaires from 561 children attending for paediatric sleep studies at GOSH were reviewed (157 retrospectively and 404 prospectively). Diagnosis was highly variable, but patients with neuromuscular disorders (NMDs) and craniofacial anomalies were best represented with, respectively, 104 and 58 patients in each disease group. Smaller groups included patients with T21 (n = 35), patients with a metabolic condition (n = 32) and those with a Hb (n = 20). The "other" group (n = 157) included children with conditions that were not common enough to be considered as a separate group. This includes a wide variety of conditions, including Prader–Willi syndrome (n = 10), other chromosome microdeletions with no specific name (n = 6), achondroplasia (n = 3), VACTERL (n = 4), Beckwith–Wiedemann syndrome (n = 3), and Escobar syndrome (n = 2), amongst others. We refer to Table 1 for a detailed list of conditions. An additional 155 children referred for assessment of SDB with no other chronic co-morbidities were also assessed. These additional patients were predominantly referred by ear, nose and throat (ENT) surgeons.

The age range of patients in different disease groups is summarised in Table 2. Overall, the median age of our study population was 7 years (range 2–19 years), but neuromuscular patients were older with a median age of 12 years (p < .0001). Separating the retrospective and prospective data had no impact on any of the baseline characteristics (Table 3).

3.2 | Polysomnography and cardiorespiratory polygraphy

From all sleep studies, 523 were conducted in air, 37 on NIV (14 on continuous positive airway pressure ventilation and 23 on bi-level positive airway pressure ventilation) and one with nasopharyngeal airway in situ. Overall, the median PSQ score was 0.42 (range 0.0–1.0), and 353 patients (67.5%) had a positive PSQ. The median score was lower for neuromuscular patients at 0.20 (range 0.00–0.89) and higher for patients with T21 at 0.65 (range 0.16–0.94). The median AHI was 1.5 events per hr (range 0–133); 338 patients (64.6%) had a CRP with a total AHI ≥ 1 per hr, and 126 with a total AHI ≥ 5 per hr (24.1%).

When a PSQ cut-off of 0.33 was used, the overall sensitivity for all 523 PSQs in predicting SDB (AHI ≥ 5) was 71.6% (95% confidence interval [CI] 61.4%–80.4%) and specificity 38.8% (95% CI 34%–43.4%). If AHI ≥ 1 was taken to be representative of SDB (Marcus, 1992), the sensitivity for all PSQs fell to 67.1% (95% CI 60.5%–73.3%) and specificity improved slightly to 39.8% (95% CI 34.6%–45.3%). For patients with OSA and no other underlying chronic disease, sensitivity was 76.3% when AHI ≥ 5 was used as an indicator of SDB, but this was much lower when looking specifically at neuromuscular patients (25%) or patients with T21 (36.7%).

TABLE 1 List of conditions included in the "other" group

Patient number	Diagnosis
1	Prader–Willi syndrome
2	Chromosome 18q deletion, epilepsy, cleft palate (repaired)
3	Previous submucous cleft palate (repaired)
4	Dilated cardiomyopathy
5	Foetal alcohol syndrome, cleft palate (repaired)
6	Arthrogryposis, epilepsy, perisylvian syndrome
7	Congenital cerebellar ataxia
8	Congenital central hypoventilation syndrome
9	SOTOS syndrome
10	Arthrogryposis, scoliosis
11	Prader–Willi syndrome
12	Cleft lip and palate (repaired)
13	Potocki–Luspski syndrome
14	Beckwith–Wiedemann syndrome
15	WAGR syndrome
16	Obstructive hydrocephalus, spastic dystonic motor disorder
17	Microdeletion chromosome 2, seizures, ischaemic infarct
18	CHARGE syndrome
19	U-shaped cleft palate
20	HUPRA syndrome
21	Ex-premature baby, chronic lung disease
22	Prader–Willi syndrome
23	Emanuel syndrome, cleft palate, micrognathia
24	Choanal atresia, Pierre–Robin syndrome, cleft palate (repaired)
25	Ex-premature baby, nasal deviation
26	Bardet–Biedl syndrome
27	Achondroplasia, gastro-oesophageal disease
28	Previous cerebrovascular accident
29	Marfan syndrome
30	Global developmental delay, cerebral palsy
31	Tracheocutaneous fistula (repaired)
32	Severe scoliosis
33	Epilepsy
34	MECP2 duplication syndrome
35	Four-limb cerebral palsy, spasticity secondary to hypoxic injury, global delay
36	Cervicofacial veno-lymphatic malformation
37	Four-limb cerebral palsy, kyphoscoliosis, global developmental delay
38	Severe bronchomalacia
39	Escobar syndrome, scoliosis
40	Escobar syndrome, severe obstructive lung disease, cleft palate (repaired)
41	Monosomy 13, hypertrophic cardiomyopathy
42	Cleft palate, previous pharyngoplasty
43	Haemolytic uraemic syndrome, basal ganglia strokes, global delay
44	Cerebral palsy GMFCS V
45	Cerebral palsy GMFCS V, epilepsy, hypoxic birth injury
46	Skeletal dysplasia
47	Ex-premature baby, tracheomalacia
48	Pierre–Robin syndrome, previous cleft palate (repaired)
49	Microdeletion chromosome 4q, ex-premature baby
50	Tetralogy of Fallot, severe kyphoscoliosis
51	Neurofibromatosis, severe kyphoscoliosis
52	Aromatic amino acid decarboxylase deficiency, scoliosis
53	Chromosome 6 deletion
54	Congenital diaphragmatic hernia, tracheoesophageal fistula
55	Cleft lip, cheek and palate
56	Cerebral palsy, severe scoliosis
57	ACTA1-related congenital fibre-type disproportion
58	Bronchopulmonary artery stenosis with right ventricular hypertrophy
59	Rett syndrome
60	Ex-premature baby, tracheomalacia, pulmonary hypertension
61	Ex-premature baby, subglottic stenosis, bronchopulmonary artery stenosis
62	Severe recurrent migraines
63	Hypotonia, bulbar palsy, global developmental delay
64	Pituitary microadenoma, elevated body mass index (BMI)
65	VSD, aortic stenosis
66	Cerebral palsy
67	22q11del, complex congenital heart disease
68	Rett syndrome, scoliosis
69	Ex-premature baby, developmental delay, hypotonia
70	Lennox-Gastaux, elevated body mass index
71	Pierre–Robin syndrome, cleft palate (repaired)
72	Variant neuronal ceroid lipofucinosis, epilepsy, global developmental delay
73	Rett syndrome, global developmental delay
74	Chronic lung disease, pulmonary hypertension, epilepsy
75	Ex-premature baby, cerebral palsy, hydrocephalus, epilepsy
76	Prader–Willi syndrome, scoliosis
77	Elevated BMI, hyperinsulinism, hypothyroidism
78	Pierre–Robin syndrome, cleft palate (repaired)
79	Goldenhar syndrome

(Continues)

TABLE 1 (Continued)

Patient number	Diagnosis
80	Cerebral palsy (GMFCS V), microcephaly, global developmental delay
81	Hereditary haemorrhagic telangectasia, possible arteriovenous malformation
82	Beckwith–Wiedemann syndrome
83	Prader–Willi syndrome, scoliosis
84	Chiari malformation, facial dysmorphism, behavioural difficulties
85	Ex-premature baby, epigastric hernia repair
86	Cerebellar tonsillar ectopia, motor delay
87	Focal motor seizures, right Horner's syndrome, developmental delay
88	SOTOS syndrome
89	Sickle-cell anaemia, ex-premature baby
90	Ex-premature baby, atrial septal defects, pulmonary stenosis
91	Turner mosaicism
92	Achondroplasia
93	Ex-premature baby, subglottic stenosis, microcephaly, global delay
94	Mixed cardiomyopathy, sickle-cell trait
95	Larsen syndrome, severe kyphosis, bulbar palsy
96	Micrognathia, laryngomalacia
97	Turner syndrome, scoliosis
98	Jeune syndrome
99	Prader–Willi syndrome, scoliosis
100	Achondroplasia
101	Micrognathia, cleft palate, developmental delay
102	Chromosome 16p microdeletion, elevated BMI, learning difficulties
103	Epidermolysis bullosa, previous epilepsy
104	VACTERL
105	Perinatal meconium aspiration, seizures, global developmental delay
106	Right lung hypoplasia, aortocollateral pulmonary artery
107	Cerebral palsy, epilepsy
108	Infantile spasms, microcephaly, hypotonia, global developmental delay
109	Scoliosis, hypotonia, global developmental delay
110	Lateral meningocele syndrome, VP shunt, congenital heart disease
111	Ex-premature baby, microcephaly
112	EBV-lymphoproliferative disorder, bronchiectasis, immunodeficiency
113	Tuberous sclerosis
114	Four-limb motor disorder, microcephaly, seizures, kyphosis
115	Scheuermann's disease, developmental delay

(Continues)

TABLE 1 (Continued)

Patient number	Diagnosis
116	Coeliac disease, asthma
117	Growth hormone deficiency, scoliosis
118	Prader–Willi syndrome
119	Cerebral palsy, epilepsy
120	Polymicrogyria, seizures, global developmental delay
121	Type IV laryngeal cleft, tracheobronchomalacia, previous tracheostomy
122	Pulmonary lymphangiectasia
123	Ex-premature baby, laryngomalaica, ariepiglottoplasty
124	Idiopathic intracranial hypertension
125	Ex-premature baby, hypotonia
126	Ex-premature baby, global developmental delay
127	VACTERL, repaired tracheoesophageal fistula
128	Prader–Willi syndrome
129	Fragile X, elevated BMI
130	Segmentation anomalies of upper cervical spine, scoliosis
131	Chromosome 9 deletion, dystonia, dysmorphic features, global delay
132	Prader–Willi syndrome, lumbar scoliosis
133	Diamond–Blackfan syndrome, microcephaly, global delay
134	Systemic juvenile idiopathic arthritis, previous HLH
135	Beckwith–Wiedemann syndrome
136	VACTERL
137	Ex-premature baby, chronic lung disease, pertussis in neonatal period
138	Pierre–Robin syndrome, cleft palate (repaired), previous tracheostomy
139	Congenital central hypoventilation syndrome
140	Cri-du-chat 5p deletion, autism, learning difficulties
141	Pierre–Robin syndrome, previous tracheostomy, subglottic stenosis
142	Mitochondrial disease, scoliosis, global developmental delay
143	Prader–Willi syndrome, elevated BMI
144	Ex-premature baby, chronic lung disease
145	Dravet syndrome, epilepsy, elevated BMI, learning difficulties
146	Bulbar palsy, dysphagia, unsafe swallow
147	Cerebellar tonsillar ectopia, autism, developmental delay
148	Transverse myelitis, quadriparetic
149	Ex-premature baby, global developmental delay, plagiocephaly
150	Severe laryngomalacia, previous aryepiglottoplasty
151	Submucous cleft palate, conductive hearing loss
152	Pierre–Robin syndrome, cleft palate (repaired)
153	Lowe syndrome, Fanconi anaemia, scoliosis

(Continues)

TABLE 1 (Continued)

Patient number	Diagnosis
154	VACTERL, scoliosis
155	Turner syndrome
156	Scoliosis, severe lordotic posture
157	Hypotonia, scoliosis, developmental delay

Sensitivity and specificity of the PSQ, using either AHI ≥ 1 or AHI ≥ 5 as being representative of SDB, were calculated for each individual disease group, along with PPVs and NPVs. These results are summarised in Tables 4 and 5.

3.3 | Correlation between total apnoea–hypopnoea index, obstructive apnoea–hypopnoea index and Pediatric Sleep Questionnaire

The correlation between AHI and PSQ score was calculated for all subjects and for each individual disease group. The results are summarised in Table 6. This correlation is statistically significant for the study population as a whole, although not strong with a value of $r = .11$. Correlation improved but remained weak (i.e. below 0.5) in the patients with craniofacial disorders ($r = .27$) and T21 ($r = .34$; Figure 1a,b).

The correlation between oAHI and PSQ score was also calculated for all subjects and for each individual disease group, and gave similar results. A significant correlation was found for the craniofacial and T21 sub-groups. The results are summarised in Table 7.

Finally, the correlation of AHI and oAHI was also calculated. This is shown in Table 8 and Figure 2. The correlation is similar to that described in the study by Chervin et al. (personal communication). We were concerned that children with chronic underlying medical conditions might have elevated central AHI and that this might impact significantly on the total AHI; that there is an excellent correlation between oAHI and total AHI in all groups vindicates the use of total AHI (also used in the original PSQ study; Chervin et al., 2000) as our comparative variable.

3.4 | Impact of cognitive domain on Pediatric Sleep Questionnaire

To determine the impact of the cognitive domain (questions C1–6 from PSQ) on the outcome of the PSQ score, sensitivity and specificity were assessed (with AHI ≥ 5 being indicative of SDB; Chervin et al., 2000) in otherwise healthy children referred for assessment of OSAS ($n = 69$) and those with underlying chronic medical conditions ($n = 230$) in a sub-group analysis (Table 9). This showed a significant difference in the PSQ score with and without the cognitive domain for children with underlying chronic diseases ($p < .01$), but not for those with suspected OSAS and no other co-morbidities. The sensitivity of the PSQ score was slightly improved in both subjects

TABLE 2 Baseline characteristics of different disease groups

	All ($n = 561$)	Craniofacial ($n = 58$)	NM ($n = 104$)	T21 ($n = 35$)	Metabolic ($n = 32$)	Hb ($n = 20$)	Other ($n = 157$)	OSAS ($n = 155$)
Age (years)	7 [2–19]	5.5 [2–19]	12 [2–19][a]	6 [2–16]	8 [2–17]	8 [2–13]	6 [2–18]	5 [2–18]
PSQ score	0.42 [0.0–1.0]	0.38 [0.0–1.0]	0.20 [0.0–0.89][a]	0.65 [0.16–0.94][a]	0.37 [0.07–0.76]	0.37 [0.0–0.68]	0.44 [0.0–1.0]	0.52 [0.0–0.95]
AHI	1.50 [0.0–133.0]	1.45 [0.0–133.0]	1.45 [0.0–26.8]	2.10 [0.0–36.8]	2.35 [0.0–45.2]	1.60 [0.0–28.8]	1.50 [0.0–54.7]	1.00 [0.0–41.0]
oAHI	0.40 [0.0–60.6]	0.85 [0.0–60.6]	0.10 [0.0–11.3][a]	1.50 [0–36.6][a]	0.30 [0.0–45.2]	1.25 [0.0–27.1]	0.30 [0.0–53.0]	0.40 [0.0–40.7]
SpO_2%	97 [88–99]	97 [93–98]	96 [93–99]	96 [92–99]	97 [94–99]	95 [88–99]	97 [89–99]	97 [94–99][a]
ODI	4 [0–135]	3 [0–135]	4 [0–73]	6 [0–41][a]	4 [0–53]	5 [0–35]	4 [0–56]	3 [0–42]
Mean $TcPCO_2$ (mmHg)	44 [26–67]	45 [34–58]	44 [37–60]	46 [38–55][a]	43.5 [26– 50]	42.5 [36–47]	44 [29–67]	44 [32–51]
Peak $TcPCO_2$ (mmHg)	48 [35–76]	49 [37–68]	48 [40–69]	50 [42–61]	48.5 [39–60]	45.5 [40–52][a]	48 [35–76]	48 [37–67]

All data presented as medians (range).

AHI, apnoea–hypopnoea index; Hb, haemoglobinopathy; NM, neuromuscular; oAHI, obstructive apnoea–hypopnoea index; ODI, oxygen desaturation index; OSAS, obstructive sleep apnoea syndrome; PSQ, Pediatric Sleep Questionnaire; SpO_2, oxygen saturations; T21, Trisomy 21; $TcPCO_2$, transcutaneous carbon dioxide.

[a]Indicates where median differs significantly compared with other groups (Kruskal–Wallis with Dunn's multiple comparison test).

TABLE 3 Baseline data for patients, separated into that collected retrospectively and prospectively

	Retrospective (n = 152)	Prospective (n = 409)	p[a]
Age (years)	7.5 [2.0–19]	6 [2.0–18.0]	.08
PSQ score	0.4 [0–0.90]	0.44 [0–1.0]	.17
AHI	1.3 [0–54.7]	1.6 [0–133]	.71
oAHI	0.3 [0–53.0]	0.5 [0–60.6]	.31
SpO_2%	97 [92–99]	97 [88–99]	.14
ODI	3.5 [0–97]	4 [0–135]	.86
Mean $TcPCO_2$	44 [29–67]	44 [26–63]	.88
Peak $TcPCO_2$	48 [35–76]	48 [37–69]	.93

AHI, apnoea–hypopnoea index; oAHI, obstructive apnoea–hypopnoea index; ODI, oxygen desaturation index; PSQ, Pediatric Sleep Questionnaire; SpO_2, oxygen saturations; $TcPCO_2$, transcutaneous carbon dioxide.

[a]All non-parametric *t*-tests (Mann–Whitney) as no data were normally distributed.

with only OSAS and with chronic conditions. For the OSAS group, sensitivity increased from 72.7% to 75% when the cognitive domain was removed, and from 63.3% to 64.6% for the group with chronic underlying disorders.

4 | DISCUSSION

Multiple questionnaires have been developed, but the PSQ is considered to be among the best instruments for assessing OSAS in otherwise healthy children (Spruyt & Gozal, 2011). We confirm that the PSQ is a sensitive screening tool in children that only have OSA symptoms, although sensitivity and particularly specificity are lower than previously reported by Chervin et al. (2000). Compared with the original sensitivity of 85% and specificity of 87%, we describe in our study a sensitivity of 76.3% and specificity of 18.8% for the PSQ in predicting OSA as defined by CRP using an AHI of ≥ 5 as cut-off. Similar figures (sensitivity of 77.3% and specificity of 18%) were found when a cut-off AHI ≥1 was used, which, in children, is considered the limit of normality (Marcus, Omlin, Basinki, & Bailey, 1992). These sensitivity figures are in line with the paper published by the same group in 2007 that revalidated the PSQ (Chervin et al., 2007). Although that paper focused on children aged 5–12 years, the sensitivity was then shown to be 78% and the specificity 72%. In contrast with previous studies, we used an abbreviated CRP without neurophysiology measures (Jacob et al., 1995) and without oesophageal pressure probe. We therefore probably underestimated the "true score" in comparison with a full PSG with electroencephalographic leads because

TABLE 4 Sensitivity, specificity, PPV and NPV of PSQ using AHI ≥ 1 as being representative of SDB

Disease group	Sensitivity % [95% CI]	Specificity % [95% CI]	PPV % [95% CI]	NPV % [95% CI]
All (n = 561)	67.1 [60.5–73.3]	39.8 [34.6–45.3]	42.2 [37.0–47.6]	64.9 [58.0–71.4]
Craniofacial (n = 58)	60.0 [38.7–78.9]	45.5 [28.1–63.7]	45.5 [28.1–63.7]	60.0 [38.7–78.9]
NM (n = 104)	36.7 [19.9–56.1]	75.7 [64.3–84.9]	37.9 [20.7–57.7]	74.7 [63.3–84.0]
T21 (n = 35)	84.0 [63.9–95.5]	20.0 [2.5–55.6]	72.4 [52.8–87.3]	33.3 [4.3–77.7]
Metabolic (n = 32)	53.9 [25.1–80.8]	42.1 [20.3–66.5]	38.9 [17.3–64.3]	57.1 [28.9–82.3]
Hb (n = 20)	66.7 [34.9–90.1]	50.0 [15.7–84.3]	66.7 [34.9–90.1]	50.0 [15.7–84.3]
Other (n = 157)	70.6 [56.2–82.5]	32.1 [23.3–41.8]	33.3 [24.6–43.1]	69.4 [54.6–81.2]
OSA (n = 155)	77.3 [65.3–86.7]	18.0 [10.6–27.6]	41.1 [32.4–50.3]	51.6 [33.1–69.9]

CI, confidence interval; Hb, haemoglobinopathy; NM, neuromuscular; NPV, negative predictive value; OSA, obstructive sleep apnoea; PPV, positive predictive value; T21, Trisomy 21.

TABLE 5 Sensitivity, specificity, PPV and NPV of PSQ using AHI ≥ 5 as being representative of SDB

Disease group	Sensitivity % [95% CI]	Specificity % [95% CI]	PPV % [95% CI]	NPV % [95% CI]
All (n = 561)	71.6 [61.4–80.4]	38.8 [34.4–43.4]	14.4 [8.9–21.6]	88.8 [80.1–94.3]
Craniofacial (n = 58)	55.6 [21.2–86.3]	42.9 [28.8–57.8]	15.2 [5.1–31.9]	84.0 [63.9–95.5]
NM (n = 104)	25.0 [0.63–80.6]	72 [62.1–80.5]	3.5 [0.09–17.8]	96.0 [88.8–99.2]
T21 (n = 35)	36.7 [19.9– 56.1]	75.7 [64.3–84.9]	37.9 [20.7–57.7]	74.7 [63.3–84.0]
Metabolic (n = 32)	50.0 [11.8–88.2]	42.3 [23.4–63.1]	16.7 [3.6–41.4]	78.6 [49.2–95.3]
Hb (n = 20)	40.0 [5.3–85.3]	33.3 [11.8–61.6]	16.7 [2.1–48.4]	62.5 [24.5–91.5]
Other (n = 157)	80.0 [59.3–93.2]	33.3 [25.4–42.1]	18.5 [11.7–27.1]	89.8 [77.8–96.6]
OSA (n = 155)	76.3 [59.8–88.6]	18.8 [12.2–27.1]	23.4 [16.3–31.8]	71.0 [52.0–85.8]

CI, confidence interval; Hb, haemoglobinopathy; NM, neuromuscular; NPV, negative predictive value; OSA, obstructive sleep apnoea; PPV, positive predictive value; T21, Trisomy 21.

TABLE 6 Correlation of AHI with PSQ score

Disease group	Spearman *r* [95% CI]	*p*
All (*n* = 561)	0.11 [0.02–0.19]	.009[a]
Craniofacial (*n* = 58)	0.27 [0.00–0.50]	.044[a]
NM (*n* = 104)	−0.02 [−0.22–0.18]	.821
T21 (*n* = 35)	0.34 [0.00–0.61]	.044[a]
Metabolic (*n* = 32)	0.20 [−0.17–0.51]	.281
Hb (*n* = 20)	0.01 [−0.44–0.46]	.961
Other (*n* = 157)	0.10 [−0.06–0.25]	.228
OSA (*n* = 155)	0.09 [−0.08–0.25]	.280

CI, confidence interval; Hb, haemoglobinopathy; NM, neuromuscular; OSA, obstructive sleep apnoea; T21, Trisomy 21.
[a]Indicates where the correlation of total AHI with PSQ score was statistically significant.

TABLE 7 Correlation of obstructive AHI with PSQ score

Disease group	Spearman *r* [95% CI]	*p*
All (*n* = 561)	0.20 [0.12–0.28]	< .001[a]
Craniofacial (*n* = 58)	0.35 [0.09–0.56]	.007[a]
NM (*n* = 104)	0.15 [−0.04–0.34]	.123
T21 (*n* = 35)	0.46 [0.14–0.69]	.006[a]
Metabolic (*n* = 32)	0.33 [−0.04–0.61]	.069
Hb (*n* = 20)	0.05 [−0.41–0.49]	.828
Other (*n* = 157)	0.12 [−0.04–0.27]	.140
OSA (*n* = 155)	0.06 [−0.10–0.22]	.446

CI, confidence interval; Hb, haemoglobinopathy; NM, neuromuscular; OSA, obstructive sleep apnoea; T21, Trisomy 21.
[a]Indicates where the correlation of obstructive AHI (oAHI) with PSQ score was statistically significant.

hypopnoeas causing arousals or awakenings cannot be scored. This is especially true for children with OSA that have predominantly hypopnoeas that cause arousals only.

This is to our knowledge the largest cohort of patients to show that the PSQ is less useful in children with chronic disease especially when a cut-off value AHI ≥5 is used, most notably in children with T21 and NMDs, but also in children with craniofacial anomalies or metabolic conditions, where the aetiology of SDB is more likely to be multifactorial. In line with our findings, Cielo et al. (2014) showed that the PSQ is not a good screening tool for OSAS in children with craniofacial conditions; this was a retrospective chart review of 67 children aged 2–18 years. They showed that the sensitivity of the PSQ for detecting OSAS using a cut-off AHI of ≥ 5 in craniofacial patients was 57% and specificity 48%.

The correlation between PSQ score and total AHI/oAHI is statistically significant across the entire group. However, when breaking down into groups, individual correlations are poor, with the exception of T21. This potentially reflects that children with T21 have lots of symptoms leading to high scores, whereas those with NMD report relatively few symptoms for their levels of AHI. The absent association between the PSQ score and CRP findings in these medical conditions may be attributed to symptoms being different from otherwise healthy children for which the PSQ was validated, as well as parental symptom perception that may be different. Because PSG is routinely used as a screening tool for early stages of SDB/OSAS in these at-risk populations, there is a potential bias towards less symptomatic patients. However, sensitivities of the PSQ improved (up to 84% in T21 and 36% in NMD) when using a cut-off AHI value of ≥ 1 being indicative of SDB rather than ≥ 5 in patients with underlying chronic disorders, whereas sensitivities in the OSA group remained almost unaffected (77.3% versus 76.3%).

Our findings suggest that there is a need for either disease-specific questionnaires or revalidation of the PSQ in these groups,

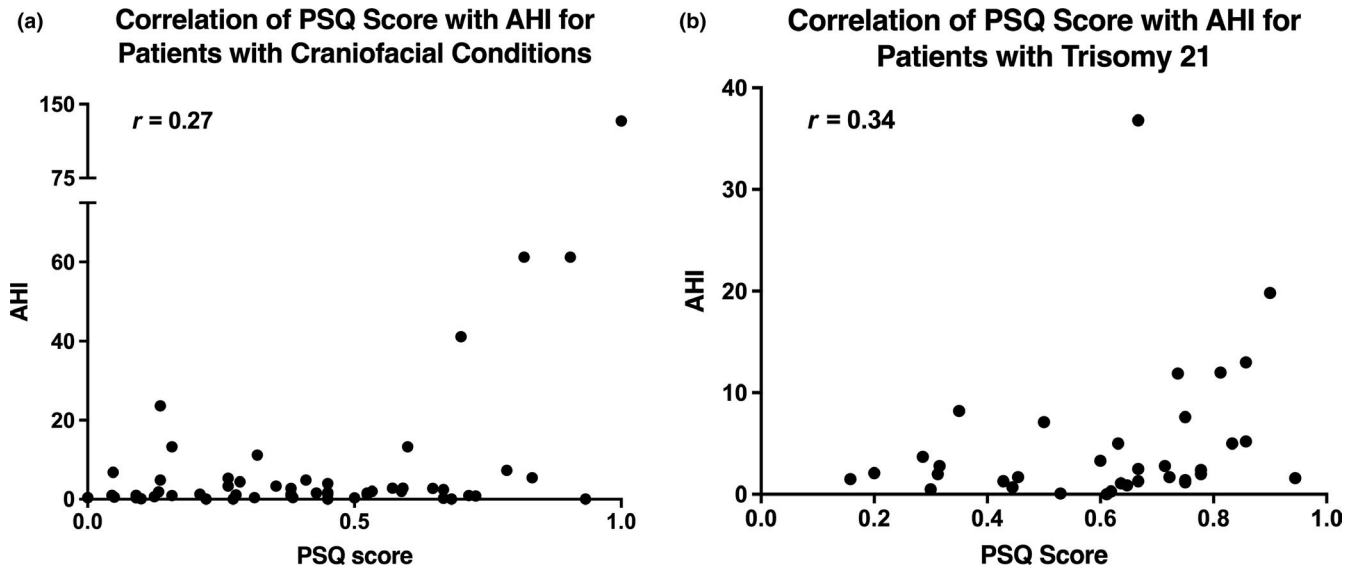

FIGURE 1 (a) Correlation of Pediatric Sleep Questionnaire (PSQ) score with apnoea–hypopnoea index (AHI) for patients with craniofacial conditions. (b) Correlation of PSQ score with AHI for patients with Trisomy 21. Whilst these correlations are statistically significant, they are weak (*r* = <0.5)

TABLE 8 Correlation of AHI and oAHI

Disease group	Spearman *r* [95% CI]	*p*
All (*n* = 561)	0.78 [0.74–0.81]	< .0001[a]
Craniofacial (*n* = 58)	0.86 [0.77–0.92]	< .0001[a]
NM (*n* = 104)	0.60 [0.45–0.71]	< .0001[a]
T21 (*n* = 35)	0.96 [0.93–0.98]	< .0001[a]
Metabolic (*n* = 32)	0.75 [0.53–0.87]	< .0001[a]
Hb (*n* = 20)	0.97 [0.93–0.99]	< .0001[a]
Other (*n* = 157)	0.68 [0.56–0.76]	< .0001[a]
OSA (*n* = 155)	0.90 [0.86–0.92]	< .0001[a]

CI, confidence interval; Hb, haemoglobinopathy; NM, neuromuscular; OSA, obstructive sleep apnoea; T21, Trisomy 21.

[a]Indicates where the correlation of total AHI and obstructive AHI (oAHI) is statistically significant (all $p < .0001$ in this analysis).

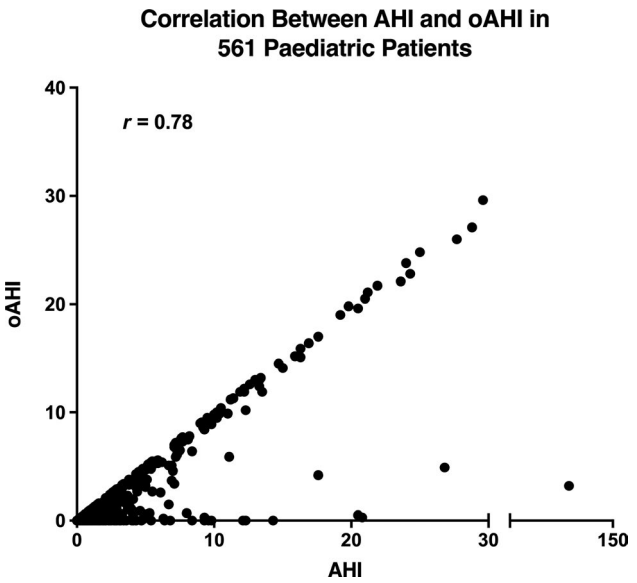

FIGURE 2 Correlation between apnoea–hypopnoea index (AHI) and obstructive (o)AHI in 561 paediatric patients

perhaps leading to a different cut-off score to 0.33. A disease-specific questionnaire has been developed to screen for SDB in patients with NMD that includes symptoms of diaphragmatic weakness, but this questionnaire has only been validated in adults (Steier et al., 2011). Similarly, a specific 14-item questionnaire was proposed as a screening tool for OSA in children with T21 from infancy to 6 years (Sanders, Hill, Evans, & Tuffrey, 2015), but has not yet been fully validated. Alternative screening modalities may be more appropriate for OSAS in children with complex underlying disorders. Most modalities including audio/video recordings or nocturnal oximetry have not been sufficiently studied in children with the complex medical disorders that are represented in this study. In children and adolescents with NMD, SDB was predicted from simple daytime respiratory function tests (Mellies et al., 2003), but data from Bersanini et al. could not confirm these findings (Bersanini et al., 2012).

As cognitive symptoms in these patients are likely to be related to their complex medical conditions rather than be a consequence of OSAS, we performed a sub-analysis on our cohort after removing the cognitive domain of PSQ in an attempt to see whether this might affect sensitivity and specificity in this group of patients. This post hoc sub-analysis showed a significant difference in the PSQ score with and without the cognitive domain included for children with underlying chronic diseases ($p < .01$), but not for those simply with suspected OSAS and no other co-morbidities. In both groups, sensitivity was slightly improved without the cognitive domain, although specificity was reduced; this suggests that removal of the cognitive items may increase sensitivity of the PSQ in detecting OSAS in all children. However, as this is a post hoc analysis on a subset of patients, albeit a large cohort (*n* = 299) with data collected prospectively, future studies are needed to prospectively validate new questionnaires that both include and exclude the symptoms related to the cognitive domain in an independent cohort of patients with complex disorders and with simple OSAS.

One potential limitation to this study comes from the use of an abbreviated PSG without neurophysiology measures. The scoring used adapted rules as per the 2012 AASM guidelines for the scoring of sleep and associated events (Berry & Gamaldo, 2012). This abbreviated CRP without electroencephalographic, electrooculographic or electromyographic leads has, however, previously been demonstrated to be an accurate tool for the detection of OSAS (Alonso Álvarez et al., 2008; Jacob et al., 1995). In addition, no oesophageal pressure probe was used in our study, again in contrast to the original paper (Chervin et al., 2000). But CO_2 measurements were similar across the study population (Table 2).

TABLE 9 Impact of cognitive domain on sensitivity and specificity of PSQ for children with chronic underlying conditions and those referred for assessment of OSAS

Disease group	Median PSQ score With cognitive domain (range)	Median PSQ score without cognitive domain (range)	*p*	Sensitivity [95% CI] and specificity [95% CI] with cognitive domain	Sensitivity [95% CI] and specificity [95% CI] without cognitive domain
OSAS (*n* = 69)	0.53 [0.05–0.82]	0.54 [0.0–0.81]	.359	72.7 [49.8–89.3] 19.2 [9.2–33.3]	75.0 [57.8–87.9] 18.2 [7.0–35.5]
Other (*n* = 230)	0.38 [0.0–0.95]	0.37 [0.0–1.0]	<.01	63.3 [43.9–80.1] 43.5 [36.5–50.7]	64.5 [45.4–80.8] 46.2 [39.2–53.4]

CI, confidence interval; OSAS, obstructive sleep apnoea syndrome; PSQ, Pediatric Sleep Questionnaire.

Another limitation to the present study is the lack of prospectively collected individual and socio-demographic data, including gender, language and BMI. Neuromuscular patients as a group were older (median age 12 years) compared with the other sub-groups. This can potentially affect the resulting outcome as OSAS shows an age distribution with a peak prevalence occurring at 2–8 years of age, coinciding with the peak age of tonsillar and adenoidal hypertrophy (DelRosso, 2016). Male gender, socioeconomic status and obesity are known risk factors for SDB in children (Chervin et al., 2003; Kohler et al., 2009; Redline et al., 1999; Spilsbury et al., 2006; Valipour, 2012), but these were not systematically recorded in this study. This potentially creates a bias in the results. In addition, language difficulties can affect the understanding of the questionnaire and the quality of the data. This, again, could impact the final PSQ scores and is especially true given the international population seen at GOSH.

In conclusion, the sensitivity, specificity, and PPVs of the PSQ were all low in children with complex underlying disorders when a cut-off AHI of ≥ 5 is used. The PSQ is therefore not a good screening tool for OSAS in these children although, as NPVs were good, it has some merit in ruling out sleep disordered breathing. More research is needed to develop better screening tools in order to be able to detect those patients that need to undergo the gold-standard PSG.

ACKNOWLEDGEMENTS

Special thanks to the Sleep Team, who contributed greatly to the analyses of the sleep cardiorespiratory polygraphies, and to all the children and their parents. The institution where the work was performed: Great Ormond Street Hospital for Children, NHS Foundation Trust.

FUNDING INFORMATION

This research did not receive any specific grant from funding agencies in the public, commercial or not-for-profit sectors.

CONFLICT OF INTEREST

None.

AUTHOR CONTRIBUTIONS

All authors have seen and approved the manuscript. RP performed the data collection, did the statistical analysis and helped write the article; CG helped in the data collection and wrote the article. AL helped in the data collection and critical evaluation of the article. KR and MS helped in the study design and critical evaluation of the article. FA had a critical evaluation of the article.

ORCID

Kylie Russo https://orcid.org/0000-0001-8426-0042
Martin Samuels https://orcid.org/0000-0002-9811-8335

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How to cite this article: Pabary R, Goubau C, Russo K, Laverty A, Abel F, Samuels M. Screening for sleep-disordered breathing with Pediatric Sleep Questionnaire in children with underlying conditions. *J Sleep Res*. 2019;28:e12826. https://doi.org/10.1111/jsr.12826