International Pediatric Otolaryngology Group (IPOG) consensus recommendations : evaluation and management of congenital tracheal stenosis

International Pediatric Otolaryngology group (IPOG) consensus on the diagnosis and management of pediatric obstructive sleep apnea (OSA).

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Conflict of interest: none

Abstract:

Objective: To develop an expert-based consensus of recommendations for the diagnosis and management of pediatric obstructive sleep apnea

Methods: A two-iterative Delphi method questionnaire was used to formulate expert recommendations by the members of the International Pediatric Otolaryngology Group (IPOG)

Results: Twenty-six members completed the survey. Consensus recommendations (> 90% agreement) are formulated for 15 different items related to the clinical evaluation, diagnosis, treatment, postoperative management and follow-up of children with OSA.

Conclusion: The recommendations formulated in this IPOG consensus statement may be used along with existing clinical practice guidelines to improve the quality of care and to reduce variation in care for children with OSA.

Key words: adenotonsillectomy, endoscopy, pediatric, polysomnography, obstructive sleep apnea, treatment

Consensus objectives

Pediatric obstructive sleep apnea (OSA) is a common condition that affects 2% to 4% of the population and can result in long term morbidity including adverse neurocognitive and cardiovascular outcomes. Adenotonsillar hypertrophy is the most common cause of OSA in children and adenotonsillectomy (ATE) is the first line treatment.

The prevalence of OSA is much higher in children with comorbidities such as Down syndrome, genetic and craniofacial disorders and obesity. In these children, the etiology of this sleep disorder is multifactorial, often involving multilevel upper airway obstruction and resulting in persistent OSA after ATE.

Recently, there has been increasing awareness among physicians and otolaryngologists of the importance of early recognition and treatment of OSA. There is also an evolution in recognition of the importance, and the need for a multidisciplinary approach for children with comorbidities who are likely to have persistent OSA following ATE. Despite the availability of guidelines and recommendations published in peer reviewed journals on the diagnosis and treatment of pediatric OSA, there is still a large variability in clinical practice which offers an opportunity for quality improvements. [1], [2], [3], [4],[5], [6, 7]

The aim of this paper is to develop an international expert-based consensus of recommendations for the diagnosis and management of pediatric OSA. The goal is to improve the quality of care and to reduce variation in care for children with OSA.

Target population

Infants and children with OSA, including those with underlying conditions predisposing them to upper airway obstruction.

Intended users

These consensus recommendations are targeted towards: otolaryngologists, pediatricians, sleep medicine physicians, primary care providers and other health care professionals taking care of children with obstructive sleep apnea.

Methods

The mission of the International Pediatric Otolaryngology Group (IPOG) is to develop expert-based consensus recommendations for the management of pediatric otolaryngologic disorders with the goal of improving patient care.

Two iterations of Delphi method questionnaires were used to formulate expert recommendations on the diagnosis and management of pediatric OSA. An online survey was designed by 3 authors of the group (PB, AB, SP). This survey was sent out to all members of IPOG and responses were collected after a 4 weeks period with reminders sent to enhance response rate. Based on the responses of the first round and the comments of the responders, a second round of questions was designed and answers were collected over another 4 weeks' period. The combined results of these 2 rounds were analyzed by BP and AB and all (26) IPOG respondents contributed to the preparation of the manuscript. A critical and independent review of the final manuscript was done by MC. A consensus recommendation is reached when there is > 90% agreement among the authors on a survey question.

This consensus recommendation represents the eighth publication by the group.

Abbreviations

AHI: apnea/hypopnea index

AASM: American Academy of Sleep Medicine

AAO-HNSF: American Academy of Otolaryngology Head-Neck Surgery Foundation

ATE: adenotonsillectomy

CPAP: continuous positive airway pressure

DISE: drug induced sleep endoscopy

MRI: magnetic resonance imaging

oAHI: obstructive apnea/hypopnea index

OSA: obstructive sleep apnea

oSDB: obstructive sleep disordered breathing

PSG: polysomnography

PSQ-SDB: pediatric sleep questionnaire – sleep disordered breathing

Disclaimer

Members of the International Pediatric ORL group (IPOG) prepared this report. Consensus recommendations are based on the collective opinion of the members of this group. Any person seeking to apply the report in their clinical practice is expected to use independent medical judgment in the context of patient and institutional circumstances.

Recommendations and justifications. The recommendations are outlined in the following subheadings:

1. Definitions

2. Diagnostic considerations

3. Treatment

4. Follow-up

1. Definitions

We used the definitions of American Academy of Otolaryngology Head-Neck Surgery Foundation (AAO-HNSF) [4] as follows:

Obstructive sleep disordered breathing (oSDB) is a clinical diagnosis that includes a clinical spectrum ranging from snoring to obstructive sleep apnea.

Obstructive sleep apnea (OSA) = oSDB with an abnormal polysomnography with an obstructive apnea/hypopnea index (oAHI) > 1 event/hour. This definition implies that a diagnosis of OSA cannot be established without objective testing i.e. PSG.

OSA severity is categorized according to Kaditis et al. [2]

Primary snoring = snoring > 3 nights a week with normal a PSG with an AHI <1 event/hour.

Mild OSA: oAHI >1 up to 5 events/hour

Moderate OSA: oAHI ≥ 5 up to 10 events/hour

Severe OSA: oAHI ≥ 10/hr

*Obesity i*s defined as a as BMI z-score ≥ 2 or BMI percentile > 95 percent [8]

2. Diagnostic considerations

2.1 Questionnaires. Several questionnaires have been developed as a screening tool for oSDB or to measure its impact on quality of life. The Pediatric Sleep Questionnaire – Sleep Related Breathing Disorder (PSQ-SRDB) has been used widely. It was initially designed to identify oSDB associated symptoms in clinical research when PSG was not feasible [9]. A PSQ-SRDB score $\geq$ 0.33 has 85% sensitivity and 87% specifity for predicting moderate- to-severe OSA (AHI $\geq$ 5 events/hour).

The PSQ-SRDB score is associated with baseline behavioral impairment, sleepiness and reduced quality of life, while change in score is predictive of an improvement in these parameters after ATE [10].

A consensus recommendation (92%) was reached that a validated questionnaire assessing oSDB-related symptoms should be part of the clinical evaluation in children with signs and symptoms suggestive of OSA. However, in clinical practice, most of the experts (85%) do not use such a validated questionnaire. This discrepancy opens the way for future research to define which questionnaire is most useful in clinical practice.

2.2 Clinical findings. Reporting clinical findings in a standardized way may improve communication between clinicians and may be a useful tool for follow-up. Scales have been developed for reporting clinical findings in children with symptoms of oSDB such as the Brodsky tonsil score (tonsillar hypertrophy), the Mallampati score (tongue position), and other scoring systems assessing several craniofacial features

such as the sleep clinical record [11]. Although most widely used, the Brodsky tonsil score is a poor predictor of OSA severity or site of obstruction[12] [13]. While it was acknowledged that awake flexible endoscopy may be helpful in assessing sites of obstruction such as adenoid hypertrophy or lingual tonsil hypertrophy it has been shown not to correlate with DISE findings. [14]

There was a consensus for standardized reporting of physical examination findings in children with symptoms of oSDB. Although the Brodsky scale is a poor predictor for OSA severity, a vast majority of the experts (96%) routinely use it. More than two-thirds of respondents (77%) evaluate the presence of malocclusion (malocclusion type 1-2-3, crossbite) and 44% use the modified Mallampati score.

There was no consensus regarding the use of awake flexible endoscopy with 65.4% of the experts using this as a first line diagnostic method.

2.3 Assessment of comorbid conditions. OSA may be associated with impaired quality of life, enuresis, behavioral problems and long term neurocognitive consequences such as neurocognitive delays, cardiovascular morbidity and growth failure [2], [15]. The development of these long-term adverse consequences is likely multifactorial in origin and may vary in clinical expression with similar degrees of OSA [16]. Due to the lack of longitudinal cohort studies, little is known about incidence, severity and predictors of long-term disease complications.

According to most experts (80%) a measure of quality of life should be part of the clinical evaluation in children, but only 31% apply it to their practice.

There was no consensus regarding the indications for a referral to a pediatric subspecialist (pediatric cardiologist, pediatric neurologist and/or psychologist) for assessment of cardiovascular and neurocognitive comorbid conditions (Table 1). According to most experts (84.6%) in cases of mild-to-moderate OSA, there is no need to refer the patient to a pediatric subspecialist.

In the children with severe OSA, 77% refer to a pediatric cardiologist, but only 54% feel that neurocognitive and behavioral assessment are needed.

2.4 Objective testing. Full night PSG is the gold standard for diagnosing OSA. Some guidelines such as the American Academy of Sleep Medicine (AASM) recommend PSG in all children before adenotonsillectomy (ATE) [17]. The AAO-HNSF published a clinical practice guideline with alternate indications for PSG [18] that include: 1) children with certain complex medical conditions such as obesity, Down syndrome, craniofacial abnormalities, neuromuscular disorders, sickle cell disease, or mucopolysaccharidoses. The goal of PSG in these children is to confirm the presence of obstructive events that may benefit from surgery, to define OSA severity and assist in pre-operative planning and, to obtain a baseline for comparison after surgery. PSG is also recommended in children without any of the comorbidities listed in (1) for whom the need for surgery is uncertain or when there is discordance between tonsillar size on physical examination and the reported severity of sleep-disordered breathing. However, a recent survey of clinical practice found that, many pediatric otolaryngologists do not adhere to this guideline. [5] In cases where PSG is not available, other diagnostic tools such as polygraphy or oximetry may be considered [2].

A consensus recommendation was achieved that full night PSG or in-hospital polygraphy (attended polygraphy) is the preferred method for objective testing in children with oSDB symptoms.

There was also consensus (100%) that children who are under 2 years, have Down syndrome, or have a craniofacial malformation require preoperative PSG. Ninety-six percent of experts additionally recommended pre-operative PSG in obese children. Sixty-nine percent of IPOG experts do not recommend PSG before ATE in children > 2 years without comorbidities.

2.5 Multidisciplinary teams

The care of OSA in children with obesity, Down syndrome, craniofacial malformations or neurological conditions should be integrated into a multidisciplinary team management approach involving different pediatric subspecialties.

There was consensus (92%) that a multidisciplinary team should evaluate and manage children 1) with moderate-to- severe OSA and/underlying medical conditions; 2) children with underlying medical conditions (regardless of OSA severity). This result may be influenced by the finding that the vast majority (92%) of IPOG experts are part of a multidisciplinary team.

The experts were asked about the disciplines that should be involved in such a multidisciplinary team. There was a consensus that an otolaryngologist (100 %) and a pediatric pulmonologist (92%) should be members of the multidisciplinary team. About 2/3 (61%) of the responders indicated that a maxillofacial surgeon and/or

orthodontist also should be a part of the team, and 58% indicated that a pediatrician with expertise in sleep medicine and a sleep technician should be involved.

2.6 Upper airway evaluation

Upper airway obstruction, due to multilevel collapse, has been increasingly recognized as a cause of persistent OSA after ATE. In addition, the risk of multilevel collapse increases with OSA severity [19], [20]. Drug-induced sleep endoscopy (DISE) and cine Magnetic Resonance Imaging (MRI) are currently the most commonly used tools to investigate the upper airway in children with persistent OSA [21]. DISE is widely used among pediatric otolaryngologists and the indications are not limited to children with persistent OSA [7]. Limitations of DISE include the lack of consensus regarding scoring systems and the lack of agreement regarding the ideal drugs used to obtain an induced sleep state [7], [6]. Ideally, the drug should reproduce the different sleep stage including rapid eye movement (REM) sleep; preserve mechano-and chemoreceptor input to the brainstem, maintain respiratory rhythm and upper airway muscle activity; have a short onset of action and half-life and have amnestic properties [22] A complete overview of the different drugs available is out of the scope of this chapter and excellent reviews are available [22, 23]

Some studies, performed in otherwise healthy children, have shown that DISE performed prior to ATE, may change the surgical planning in about 1/3 to 1/4 of the patients [24], [25] with comparable treatment outcomes [25]. However, many clinicians believe that the need to routinely perform an upper airway evaluation may depend upon the associated comorbidity as shown in Figure 1.

There was consensus regarding the necessity of routine airway evaluation in patients with craniofacial malformation (92%). In other subgroups such as children with Down syndrome and young children (<2 years of age), 88% and 69 % of experts, respectfully, recommend routine airway evaluation prior to ATE. In otherwise healthy children 54% of the responders recommend these evaluations.

Most IPOG members (85%) perform DISE, 19 % request cine MRIs, and 42% utilize lateral plain radiographs..

There was consensus that observations during DISE should be reported in a standardized way to allow for comparison and sharing data between clinicians. All (100%) of the experts recommended that a DISE protocol should include information as to the site and the degree of UA obstruction and 92% recommend that the drug used during DISE should be mentioned in the protocol.

There was no consensus regarding the choice of drugs used for DISE. The following drugs were used: propofol (58%), dexmedetomidine (50%) and inhalational agents (31%).

There was consensus on the following indications for DISE: suspected tongue base obstruction (96%), severe OSA in infants without laryngomalacia (96%), OSA without explanatory findings (normal clinical examination or small tonsils-adenoids) (100%), suspicion of sleep-state laryngomalacia (92%), craniofacial malformations (92%), Down syndrome (92%) Most experts perform DISE in neurological disorders and in obese children (81%). DISE is performed before ATE in otherwise healthy children with moderate-to- severe OSA by 54% of the experts and in children under 2 years of age by 46%.

2.7 Orthodontic evaluation

Abnormalities in craniofacial growth such as a narrowed hard palate caused by chronic mouth breathing and abnormal tongue position are associated with snoring, increased upper airway resistance and OSA [26].

There was consensus regarding referral to an orthodontist for children with OSA and clinical findings of malocclusion (100%) or narrow palate (92%). Though it did not achieve consensus, most experts advocate a referral to an orthodontist for patients with major craniofacial abnormalities (88%) and when persistent mouth breathing exists after treatment of nasal obstruction (77%).

3. Treatment

3.1 Treatment of mild OSA. Data from the Childhood Adenotonsillectomy Trial (CHAT) demonstrated a high rate of spontaneous resolution in children with mild OSA unless they are overweight or of African-Americans [27]. Both surgical and non-surgical treatments may be considered in children with mild OSA and an assessment of quality of life and symptom burden are helpful in such decision making [28], [29]. Most experts (81.0%) considered a trial with anti-inflammatory drugs (nasal corticosteroids/montelukast) for children with mild OSA and adenotonsillar hypertrophy as an alternative to ATE. The duration of medical treatment varied: nasal corticosteroids are used for 1 month by 24%, 3 months by 65% and up to 6 months by 11 % of experts. Montelukast is prescribed for 1 month by 31%, 3 months by 54% and up to 6 months by 15% of experts.

It should be noted that a recent U.S. Food & Drug Administration (FDA) warning regarding montelukast safety for mild disease was announced after IPOG members were surveyed (https://www.fda.gov/media/131182/download.)
This warning concerns the risk of serious neuropsychiatric adverse events with the use of montelukast in pediatric patients. FDA recommends an assessment of potential benefits and risks when prescribing montelukast for pediatric patients. We cannot exclude that respondents might have answered differently taking this warning into account.
There was consensus (100%) that children receiving medical treatment for mild OSA should be reassessed for symptomatic improvement after therapy.

3.2. **Adenotonsillectomy.** Adenotonsillectomy is the first line surgical treatment in children with OSA [30]. The majority of children improve in both subjective and objective parameters after ATE. However, objective testing shows persistent disease in 10% to 77% depending on the definition of OSA and the presence of risk factors and comorbidities [30], [31]. In otherwise healthy non-obese children, the success rate of ATE is approximately 75% [31], [32]. Risk factors for perioperative complications in children undergoing ATE have been identified and overnight inpatient postoperative monitoring is recommended after ATE for children less than 3 years age or those with severe OSA (apnea-hypopnea index ≥10 obstructive events/hour, oxygen saturation nadir <80% or both [4]). In addition, children with obesity or failure to thrive, neuromuscular, craniofacial or genetic disorders are usually observed overnight after ATE [2]. There was no consensus as to whether

postoperative monitoring should be in the pediatric intensive care unit, postoperative anesthesia unit or at the ward with oximetry.

There is a consensus regarding ATE as the first line surgical treatment for OSA in healthy children aged 2-18 years (100%). Most experts (89%) perform ATE in children under 2 years age and/or obese patients. Approximately 85% perform ATE in Down syndrome and 74% in craniofacial disorders as a first line treatment for OSA.

There was consensus that full night PSG is required preoperatively in patients with craniofacial malformation or Down syndrome. To a lesser extent, 74% of experts request PSG before surgery in children under 2 years, and 70% recommend PSG in obese patients.

Consensus was achieved among 96% of IPOG members that children under 2 years of age regardless of OSA severity, and children with Down syndrome or a craniofacial malformation and moderate-to-severe OSA be observed overnight after ATE. More than half (62%) of the respondents agreed that otherwise healthy children between 2-18 years with moderate-to- severe OSA should be observed overnight, but this did not achieve consensus. By contrast, there was consensus that otherwise healthy patients with mild OSA (oAHI <5/hr) do not need overnight observation (92%). There was no consensus for routine overnight observation for obese children with OSA. Seventy-seven percent of responders admit obese patients with moderate-to-severe OSA. Seventy-seven percent of respondents admit children overnight with mild OSA if they have Down syndrome or a craniofacial malformation

Partial (intracapsular) tonsillectomy. Intracapsular tonsillectomy consists of (partially) removing tonsillar tissue while leaving the tonsillar capsule intact. Compared to extracapsular tonsillectomy, this technique may be associated with less postoperative pain, a faster return to normal activities and oral intake and a lower risk of postoperative bleeding (<1% versus 1-3%) [33].Younger children may have a higher need for reoperation after intracapsular tonsillectomy than after total tonsillectomy [34]. Recent studies indicate that intracapsular tonsillectomy is a suitable treatment option for selected children with OSA [35], [36]. However, a lack of longitudinal data regarding intracapsular tonsillectomy precluded its applicability or inclusion in the most recent AAO-HNS tonsillectomy guidelines [37].

There was no consensus regarding intracapsular versus total tonsillectomy as the procedure of choice. Only 30% of the respondents recommended intracapsular tonsillectomy children with OSA (Table 2).

3.3 **Continuous positive airway pressure.** CPAP therapy is usually reserved for children with moderate-to-severe OSA when surgical treatment failed or if they are not considered candidates for surgical intervention [3] [38].

There was consensus that CPAP should be considered as a treatment option for the following patient categories: severe OSA (100%), patients at high risk for surgery or anesthesia (100%), obese patients (96 %) and neurologically impaired children (96%). Shared decision making is important with 88.4% of the experts indicating that they take into account the parent- and caregiver preferences when considering CPAP as a treatment option.

4. Follow-up

The outcome of treatment should be monitored subjectively (assessment of symptoms), 6 weeks and 12 months after intervention [2]. The AASM recommends PSG as the gold standard for evaluation of the response to treatment [18] but if not available, other objective measures such as polygraphy or oximetry may be considered. Objective testing may be beneficial in children with a high risk for persistent OSA (eg. severe pre-operative OSA, obesity, craniofacial malformations, Down or other syndromes, or persistent symptoms after intervention) [2].

There was no consensus regarding routine PSG after surgical treatment. Seventy-three percent of the experts only perform PSG when symptoms persist, 65.3% perform a PSG when symptoms recur (initial improvement but recurrent symptoms) and 30% routinely perform such testing always 3 months postoperatively.

There was consensus (92%) that patients with Down syndrome and craniofacial malformation should have a postoperative PSG.

An overview of all consensus statements is presented in Box 1.

5. Future perspectives

Although a consensus could be reached on 15 items regarding diagnosis and management of pediatric OSA, there are still several unsolved issues and points of disagreement. This may be partially related to local resources but may also be due to a lack of scientific evidence. Box 2 identifies subjects for future investigation to better understand and improve the care of children with OSA.

Legend to Tables and Figures

Box 1: Overview of all statements regarding the diagnosis and management of pediatric OSA for which a consensus was reached among IPOG members.

Box 2: Overview of future areas of investigation regarding the diagnosis and management of pediatric OSA.

Table 1. Response percentages to the statement: Referral to a subspecialist for assessment of comorbid conditions is required in the following subgroups.

Table 2. Response percentages to the statement: “Tonsillotomy (intracapsular partial tonsillectomy) is the preferred treatment option as opposed to tonsillectomy for the following subgroups”

Figure 1. Number of respondents to the question: “Which of the following conditions are an indication to perform DISE?”

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Table(s)

Consensus Recommendations on the diagnosis and management of pediatric OSA

1. A **validated questionnaire on oSDB symptoms** is part of the clinical evaluation in children with signs/symptoms suggestive of oSDB.
2. Findings on **physical examination** should be reported in a standardized way. The Brodsky scale should be used for assessment of tonsillar hypertrophy
3. Full night **video polysomnography and in-hospital polygraphy** (attended polygraphy) are the preferred methods for objective testing in children with oSDB symptoms
4. **Objective testing** is required in children < 2 years of age, children with craniofacial malformations including Down syndrome, and obese children
5. A **multidisciplinary team** should evaluate and manage children 1) with moderate to severe OSA and underlying medical conditions; 2) with underlying medical conditions (regardless of OSA severity). An otolaryngologist and pediatric pulmonologist are part of such a multidisciplinary team.
6. **Airway evaluation** is required in children with craniofacial malformation
7. Observations made during **DISE** should be reported in a standardized way including information on the site and degree of upper airway obstruction and the drugs used to obtain a state of induced sleep
8. **DISE** should be performed **in the following conditions**: suspicion of tongue base obstruction, severe OSA in infants without laryngomalacia, suspicion of late onset laryngomalacia, craniofacial malformations and Down syndrome
9. Children with **malocclusion or a narrow palate** should be referred to an orthodontist
10. Children receiving **medical treatment** for mild OSA should be reassessed for symptomatic improvement after therapy
11. **Adenotonsillectomy** is the first line surgical treatment for pediatric OSA in otherwise healthy children between 2-18 years.
12. Full night **polysomnography** should be performed in children with Down syndrome or craniofacial malformation **before adentonsillectomy**.
13. **Postoperative patient monitoring** should be performed in children <2 years of age and in patients with Down syndrome and other craniofacial malformations if they presented with moderate to severe OSA.
14. **Overnight monitoring is not required** in otherwise healthy patients with mild OSA
15. Taking into account parental preferences, **CPAP** is considered a treatment option in children with severe OSA high risk for anesthesia, obesity, or neurological impairment.

Table(s)

Areas for future research related to the diagnosis and management of pediatric OSA.

Is there a need for assessment of malocclusion and how could these findings be reported in a standardized way?

What is the additive value of performing awake flexible endoscopy if DISE is going to be performed at a later stage?

Which subgroup of children with OSA should be referred to cardiologist or should undergo formal neurocognitive and/or behavioral testing?

What is the role of cine MRI and lateral nasopharyngeal X -Ray as a tool for upper airway evaluation?

Is there an indication to routinely perform DISE before adenotonsillectomy regardless the age of the patient, OSA severity or underlying disease?

What is the optimal duration of medical treatment for mild OSA and what are the criteria used to define its' success or failure?

Is there a role for partial, intracapsular tonsillectomy in the management of pediatric OSA?

Table(s)

Table 1. Response percentages to the statement: “Referral to a subspecialist for assessment of comorbid conditions is required in the following subgroups.”

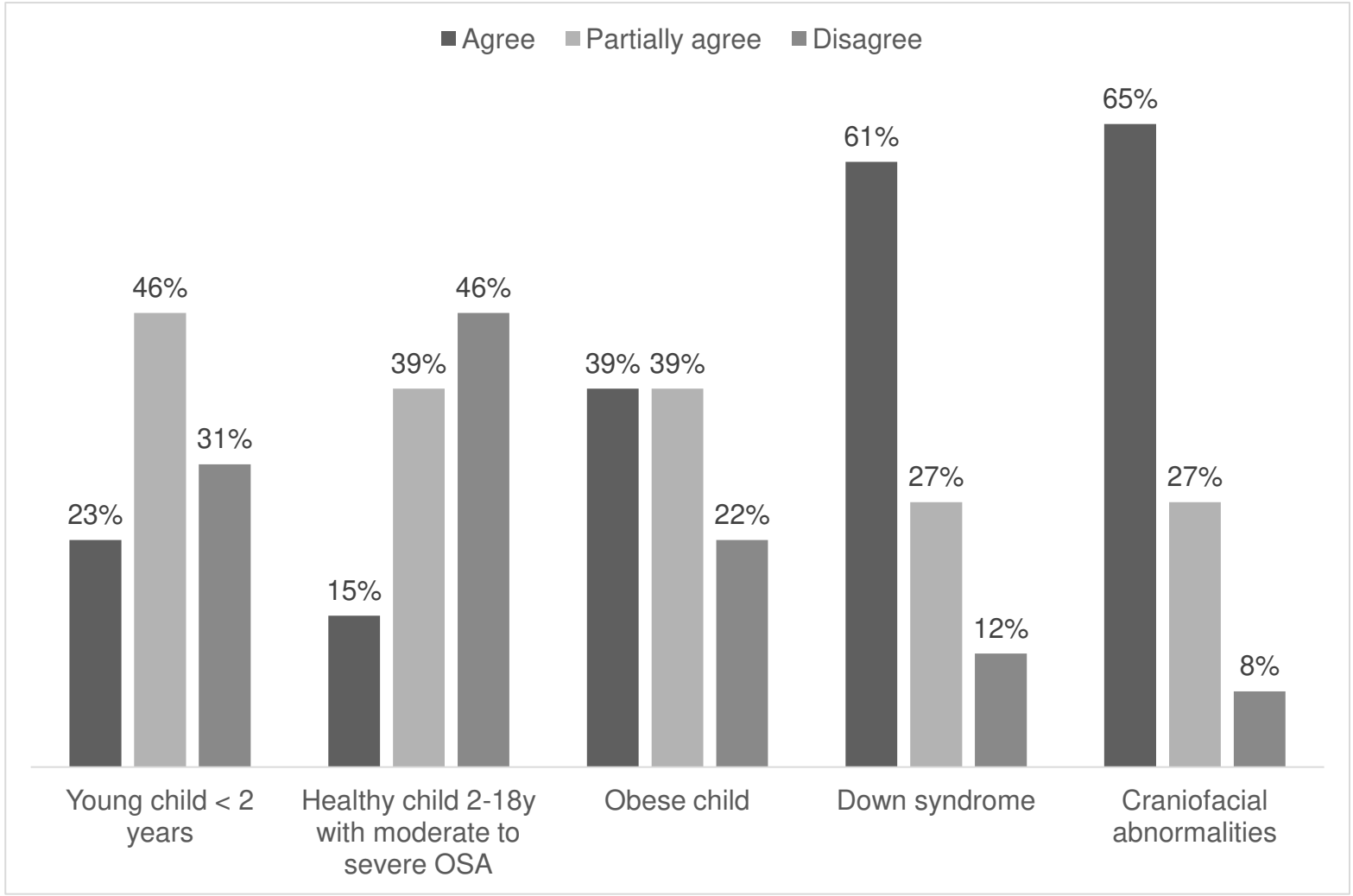
	Pediatric Cardiologist		Neurocognitive Assessment		Behavioral Assessment	
	Yes	**No**	**Yes**	**No**	**Yes**	**No**
Mild OSA	15%	85%	15%	85%	23%	77%
Moderate OSA	27%	73%	23%	77%	27%	73%
Severe OSA	77%	23%	54%	46%	54%	46%

Table(s)

Table 2. Response percentages to the statement: Tonsillotomy (intracapsular partial tonsillectomy) is the preferred treatment option as opposed to tonsillectomy for the following subgroups.

	Agree	**Disagree**
Young child < 2 years	39%	62%
Healthy child (2-18years)	31%	69%
Obese child	27%	73%
Down syndrome	23%	77%
Craniofacial abnormalities	27%	73%

Table(s)


Agree
Partially agree
Disagree
23%
46%
31%
15%
39%
46%
39%
39%
22%
61%
27%
12%
65%
27%
8%
Young child < 2 years
Healthy child 2-18y with moderate to severe OSA
Obese child
Down syndrome
Craniofacial abnormalities